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Understanding Writing Difficulties Amongst Children with Neurodevelopmental Disorders
The Cases of Dyslexia and/or Developmental Coordination Disorder (DCD)

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Understanding Writing Difficulties Amongst Children with Neurodevelopmental Disorders:
The Cases of Dyslexia and/or Developmental Coordination Disorder (DCD)

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Declaration

I hereby declare that this thesis is the results of my own investigations, except where otherwise stated. All other sources are acknowledged by bibliographic references. This work has not previously been accepted in substance for any degree and is not being concurrently submitted in candidature for any degree unless, as agreed by the University, for approved dual awards.

Yr wyf drwy hyn yn datgan mai canlyniad fy ymchwil fy hun yw'r thesis hwn, ac eithrio lle nodir yn wahanol. Caiff ffynonellau eraill eu cydnabod gan droednodiadau yn rhoi cyfeiriadau eglur. Nid yw sylwedd y gwaith hwn wedi cael ei dderbyn o'r blaen ar gyfer unrhyw radd, ac nid yw'n cael ei gyflwyno ar yr un pryd mewn ymgeisiaeth am unrhyw radd oni bai ei fod, fel y cytunwyd gan y Brifysgol, am gymwysterau deuol cymeradwy.

'A mind is a terrible thing to waste'

- **Anon**

Summary

Learning to write is onerous and takes several years to master. It is particularly taxing for children with dyslexia and/or developmental coordination disorder (DCD) who appear to have difficulties with spelling and handwriting skills which are critical for writing development. Yet, little is known about the nature of these difficulties. A complicating factor to understanding the nature of spelling and handwriting impairments in dyslexia and DCD is the reported frequent comorbidity and the unclear relationship between the two disorders.

The programmatic set of studies presented in this thesis aimed first to understand the relationship between dyslexia and DCD and the comorbidity between the two and secondly to understand the nature of spelling and handwriting impairments in dyslexia and DCD. To address these aims, the prevalence and cognitive, motor, and literacy profiles of dyslexia, DCD, and comorbid dyslexia and DCD was examined in detail. Then, the nature of handwriting difficulties in dyslexia and/or DCD was elucidated by probing profiles and correlates of handwriting in the context of fluency, legibility, and learning to form new letter-like characters.

The results demonstrated that dyslexia and DCD have independent and shared impairments and are frequently comorbid with one another. The patterns of these impairments as well as the nature of comorbidity between the two highlights the multifactorial nature of the disorders. The multifactorial nature of dyslexia and DCD also manifested in their multifaceted handwriting difficulties. Handwriting difficulties in dyslexia and DCD were apparent as dissociable impairments which reflected the nature of the specific disorder as well as impairments in early acquisition of handwriting related motor knowledge. These findings are considered in relation to implications for identification and remediation of handwriting difficulties and comorbid dyslexia and DCD.

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General Introduction Part 1: Theories of Writing: The Importance and Complexity of

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1.1. Introduction to the Thesis

Literacy skills are a uniquely human function, which are critical in modern society. Such skills surpass the boundaries of education and affect one's social integration and contribution to the economy (Fisher & Twist, 2011). At the simplest level, literacy is a tale of two halves. On the one hand, reading involves decoding written symbols of language to extract and interpret the meaning of another's ideas and thoughts. On the other hand, writing involves communicating one's own ideas and thoughts using the written symbols of language. National curricula and research have prioritised teaching and understanding how children learn to read over teaching and researching about how children learn to write. The present thesis seeks to advance the understanding of aspects of children's written productions.

Learning to convey our thoughts and ideas in written form is no easy task, taking several years to learn. For some children learning to write is taxing, especially amongst those with neurodevelopmental disorders such as dyslexia and DCD. An area of writing particularly difficult amongst children with dyslexia and DCD is in learning transcription (spelling and handwriting) skills, which are foundational for writing. Understanding the nature of transcription impairments in dyslexia and DCD is vital for developing evidence-based interventions to ameliorate difficulties and improve literacy outcomes. However, developing such an understanding is complicated by the frequent comorbidity and overlap of impairments between dyslexia and DCD, of which little is known.

Like literacy itself, this thesis presents a culmination of work that is a tale of two halves. The first half of this thesis was committed to understanding the relationship between dyslexia and DCD in terms of their cognitive, motor and literacy profiles, and the reasons for the frequent comorbidity between the two. Only after understanding the relationship between dyslexia and DCD was it possible to begin to understand the nature of transcription

(particularly handwriting) impairments in dyslexia and DCD. In the following, I bring together literatures that form the theoretical backbone of this thesis. I evaluate theories of typical skilled and developing writing before focusing on the development of spelling and handwriting skills. A second review (Chapter 2) then turns to focus on the literature pertaining to dyslexia and DCD and considers the current state of knowledge of transcription impairments in these disorders.

1.2. Theories of Writing

1.2.1. Skilled Writing: From Global to Local

Models of skilled writing are the lens from which developmentalists have and continue to view typical and atypical writing development. To date, the most influential models of skilled writing were developed by Hayes and colleagues (Chenoweth & Hayes, 2001; Hayes, 1996; Hayes, 2012; Hayes & Flower, 1980). These models followed Hayes and Flower's (1981) seminal *cognitive process model* and were chiefly concerned with describing the global processes of writing (Alamargot & Chanquoy, 2001). By focusing on global aspects of writing, models lose specificity on component aspects of written production, particularly transcription (spelling and handwriting) processes, a key topic of this thesis. Since Hayes and Flower's (1980) seminal model, other writing models have placed a greater emphasis on component, or local, aspects of writing (van Galen, 1991). Together, these models provide the foundation upon which theories of writing development are built and so they will be reviewed here briefly first.

1.2.1.1. Global models of skilled writing: Cognitive process models. Hayes and Flower (1980) proposed a model describing the cognitive processes of writing. Since its conception, this model has evolved in line with developments in the literature of writing (Hayes, 2012). Major evolutions include specifying the role of working memory (Hayes, 1996), transcription

(Chenoweth & Hayes, 2001), and attention (Hayes, 2012). The most relevant iterations of these models will be outlined.

    1.2.1.1.1. Hayes and Flowers (1980). Hayes and Flower's (1980) cognitive process model is hailed as the first attempt in delineating the cognitive processes of writing (Alamargot & Chanquoy, 2001). They were influenced by earlier work using verbal protocol analyses to examine comprehension processes (Simon & Hayes, 1976). Verbal protocol analyses require participants to verbalise all thoughts or to think aloud whilst performing a task of interest, in this case writing. Over two years, Hayes and Flowers used verbal protocol analyses of writing from a non-specified number of adults to derive their cognitive process model of writing.

    The original structure of Hayes and Flowers (1980) model is presented in Figure 1.1 (see Hayes, 1996 for a re-envisaged model). Central to this model are the *general writing processes* which receive input from *long term memory* and the *task environment.* The task environment describes external social and physical influences on the writer. Social influences include the topic of writing, the target audience, and motivational cues behind the composition. Also, in the task environment is the text so far produced by the writer to describe instances where the writer will re-read the text they have written to assist in further generating text.

    In this early version of the model, long term memory is regarded as the internal influence on the writing process. Long term memory stores several aspects of knowledge including an awareness of the audience, domain specific content knowledge, and linguistic knowledge (Hayes & Flower, 1980). The single arrow from long term memory into the planning processes in the schematic representation of the model (Figure 1.1) suggests that long term memory processes are only used in planning processes (Alamargot & Chanquoy, 2001).

The general writing process lies at the core of the model. It is composed of three sub-processes of planning, translating, and reviewing all under the control of an executive process labelled the monitor. Planning is further broken down into three sub-procedures of *generating*, *organising*, and *goal setting.* The generating procedure takes input from the task environment and long-term memory to identify stored information which will be of use in the text to be written. The information collated by the generating procedure is arranged using the organising procedure under the guidance of the aims the writer intends to fulfil (goal-setting).

The role of the translating processes is moderated by the writing plan and converts knowledge from long term memory to language. In comparison to other components of the model, Hayes and Flower (1980) are the least specific about the constituent processes required to convert knowledge into language. Specifically, the authors do not describe the processes by which pre-linguistic knowledge is transformed into language (e.g., vocabulary selection, syntax etc.) nor do they distinguish such processes from transcription (spelling and handwriting) processes. The lack of specificity in this aspect of the model probably reflects the use of verbal protocol analyses. Verbal protocol analyses rely heavily on conscious awareness of the process being described, yet the conversion of pre-linguistic ideas to strings of syntactically and grammatically correct language followed by the activation of spelling and handwriting processes is unconscious.

The authors refer to transcription skills but only in context of production in children in whom production is slower as they likely have not yet automatized these skills. In this model of skilled writing, however, it was assumed that transcription skills would be automatized and so would not impact on the writing process (Hayes, 2012). As will become clear, spelling and handwriting processes are key components of writing acquisition and production in children and continue to be important in adulthood.

In addition to planning and translating processes, reviewing is responsible for ensuring that the production matches the goals of the writing. Reviewing can either be planned (at the end of writing) or unplanned (interruption in writing) and involves the *reading* and correction (*editing*) of text produced so far in the task environment. The three general writing processes fall under the control of the monitor which acts as a controller over the selection and switching of writing processes. Flower and Hayes (1981) highlight that writers may switch between processes several times and this aspect of the model may be a source of individual differences in writers. The assumption that writers can switch between processes highlights the hierarchical nature of the model and that there is no set order for processes to become active, making the model recursive in nature.

Hayes and Flower (1980) developed their model based on verbal protocol analyses taken from an unspecified number of adults. The model was validated using the protocol from one writer who gave clear verbalisations. Using this protocol, the authors were able to delineate three types of verbalisation of goals to generate, organise, and translate. These processes map directly to aspects of the model with the same name and thus provide evidence for some aspects, but not all, of the model.

The authors recognised that the model was neither complete nor fully validated by stating that the model was "provisional, it provides a first approximate description of normal composition that can guide research and afford a valuable starting point in the search of more refined models" (Hayes & Flower, 1980, p. 10). In addition to the limitations of the model recognised by the authors, a number of specific restrictions of the model have been highlighted in several subsequent studies (see Alamargot & Chanquoy, 2001). Chief amongst the issues with this model was the lack of specificity in the translating aspect of the model, particularly in relation to spelling and handwriting (transcription) processes (Bourdin & Fayol, 1994).

Despite issues with the model – particularly in the specificity of transcription skills – Hayes and Flower's (1980) model was the first step in delineating the complex cognitive processes of written production. The model served both to stimulate studies of writing and to act as a bedrock for future studies and models of writing development (e.g., Berninger & Swanson, 1994). It is testament to the model that some of its components remain in contemporary models (Hayes, 2012).
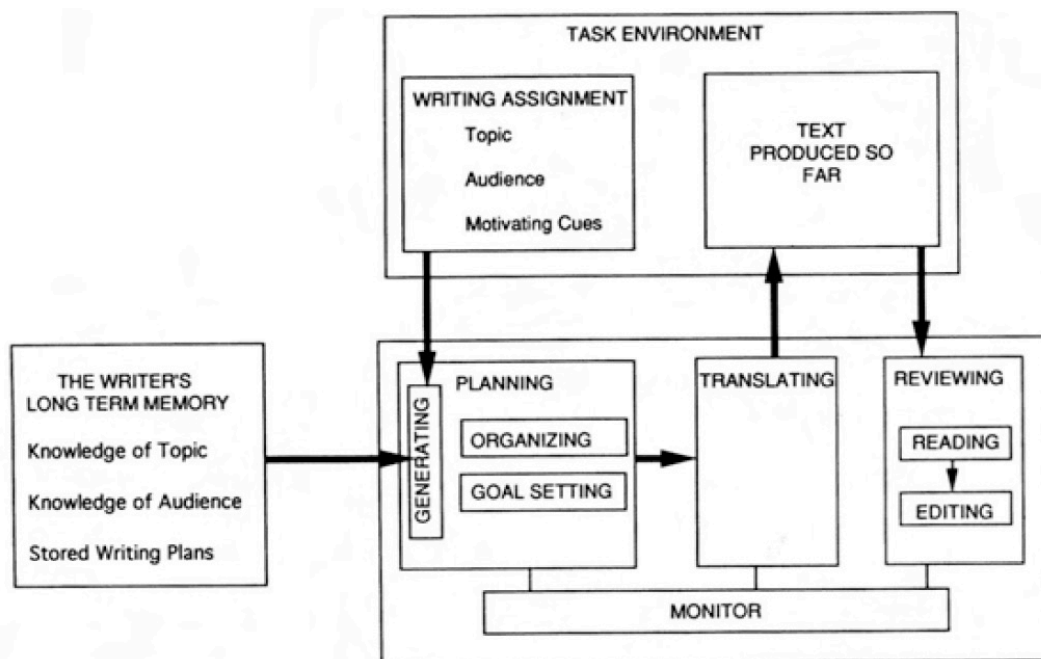
Figure 1.1. Hayes and Flower's (1980) cognitive process model of writing.

1.2.1.1.2. Chenoweth & Hayes (2001). Revisions of Hayes and Flower (1980) original model through the 1990s sought to address some of the limitations levelled at the original. This included better specifications of the writer's motivations and working memory skills (Hayes, 1996). Yet, it was not until 2001 that Chenoweth and Hayes produced a model which explicitly included transcription skills. Chenoweth and Hayes' (2001) model was an amalgamation of Hayes' (1996) update of Hayes and Flower (1980) model with contributions from (Kaufer, Hayes, & Flower, 1986) work on written sentence production. In comparison to the Hayes and Flower (1980) model, the architecture of the model changed considerably

and was defined by three distinct levels consisting of a *resource level*, *process level*, and *control level* (see Figure 1.2).

The resource level describes processes that are not specific to writing and are used by all aspects of the model. At this level, long-term memory and reading components of the original model were retained along with working memory added by Hayes (1996). Working memory was conceptualised as a limited capacity resource which contributes to all writing processes (Hayes & Chenoweth, 2006; for an alternative conceptualisation see Kellogg, 1996).

The process level distinguishes between external and internal processes. The external aspect describes processes akin to task environment from earlier models (Hayes, 1996; Hayes & Flowers, 1980). It includes both social (e.g., audience) and expanded physical (text produced and dictionaries) influences. The internal aspect of the model appears to be a considerable revision of the general processing component of Hayes and Flowers (1980) model. It is split into the four sub-procedures *proposer, translator, reviser,* and *transcriber*. The proposer receives input from multiple processes of the model (long-term memory, social, and physical influences) and appears to perform a similar task to the planning procedure in earlier models to generate ideas.

In this model, the authors provide greater specification of translator processes. They have subdivided the translating process from the original model into the *translator* and the *transcriber*. The translator is responsible for turning pre-linguistic ideas generated by the proposer into grammatical and ordered word strings. The *reviser* assesses the output from the both the *proposer* and translator to ensure the output matches the aims of the writing goals. In this sense, the reviser holds a similar role to the reviewing component of the original model.

Crucially, though, the authors specify a *transcribing* process to convert language generated by the translator to written language. During earlier models, Hayes and colleagues

(Hayes, 1996; Hayes and Flowers, 1980) had not explicitly described transcription processes, assuming spelling and handwriting processes were fully automatized in adults and so unlikely to influence written production. But work since Hayes and Flower's (1980) original model demonstrated that transcription skills constrained writing even in adults. Bourdin and Fayol (1994) found adults could recall fewer words when writing in an unfamiliar script (cursive capitals) than writing in a familiar script. Writing in an unfamiliar script was assumed to increase the cognitive cost of transcription skills and so this finding was interpreted as demonstrating that transcription skills carry some cognitive cost, even in skilled adults (Hayes, 2012; Hayes & Chenoweth, 2006).

To validate their model, Chenoweth and Hayes (2001) used verbal protocol analysis from students producing text in their native language and in a second language after three or five semesters' study. The authors specifically examined the number of words written – termed segments or bursts – demarcated by revision of the written texts and pauses (text produced without revision). Bursts demarcated by revision were hypothesised to represent instances where the proposer and translator were interrupted by the reviser whereas segments ending in pauses were believed to represent the proposer and translator not being interrupted by the reviser. Analysis of the text in conjunction with the verbal protocols showed that students wrote fewer new words per minute in their second language than in their first, made shorter production bursts in their second language, and made a larger percentage of revisions. Chenoweth and Hayes (2001) suggested that the burst size reflects translation moderated by the linguistic experience of the writer. In this view, the translator is considered a limited capacity system and where the translator's capacity is fully absorbed by effortful lexical retrieval – as in the case when learning a second language – the translator is unable to complete other functions such as applying grammar, leading to more revisions. At a more

basic level, though, this model demonstrates how linguistic processes constrain handwriting fluency.
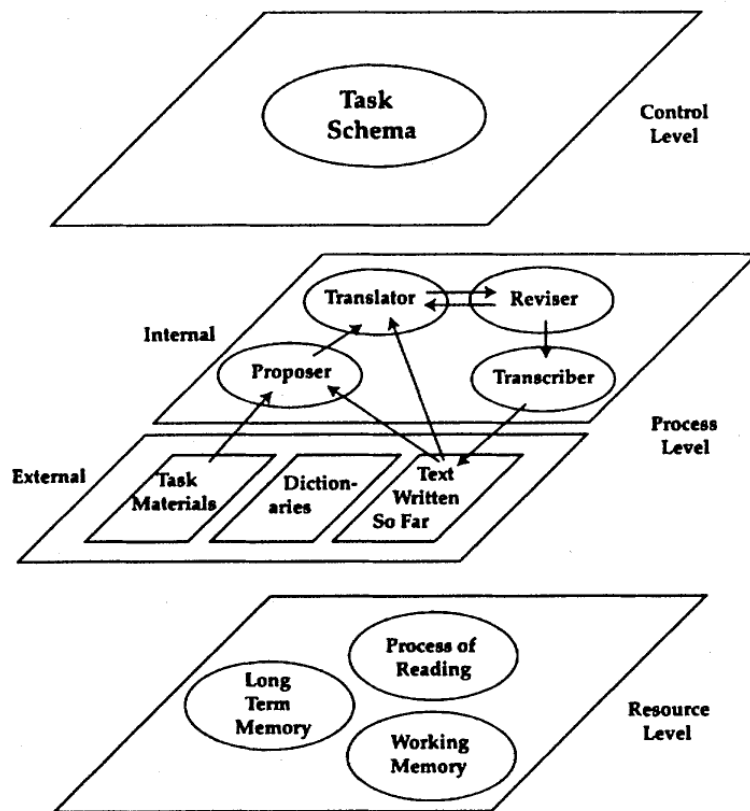
Figure 1.2. Chenoweth and Hayes (2001) three-level process model.

1.2.1.1.3. Hayes (2012). As can be seen from Figure 1.3, the overall architecture of the most recent revision of this model has changed little from Chenoweth and Hayes (2001). Notably, *attention* has been added to the resource level of the model. In the context of this model, attention is considered as the ability to attend to the relevant task and is akin to selective attention/inhibition. It is presumed to be important for the choice of writing strategy (Hayes and Berninger, 2014) but it is also likely to operate at a much lower level in the sense that attention is required to remain on the specific task of writing, particularly in the face of competing demands and when writing is effortful.

To recap, Hayes and Flowers (1980) saw their original model not as a complete work but rather "a guide to further research on writing" (Hayes & Flower, 1980, p. 29). True to the

author's wishes it has been responsible for stimulating research delineating the processes

involved in writing and has served as the foundation for developmental models of writing

(Alarmagot & Chanquoy, 2001). The model itself has undergone considerable revision since

its conception, although most of the original processes identified in the model are still present

in contemporary models. Importantly, recent iterations of the model identify transcription

skills as playing a pivotal role in written production.
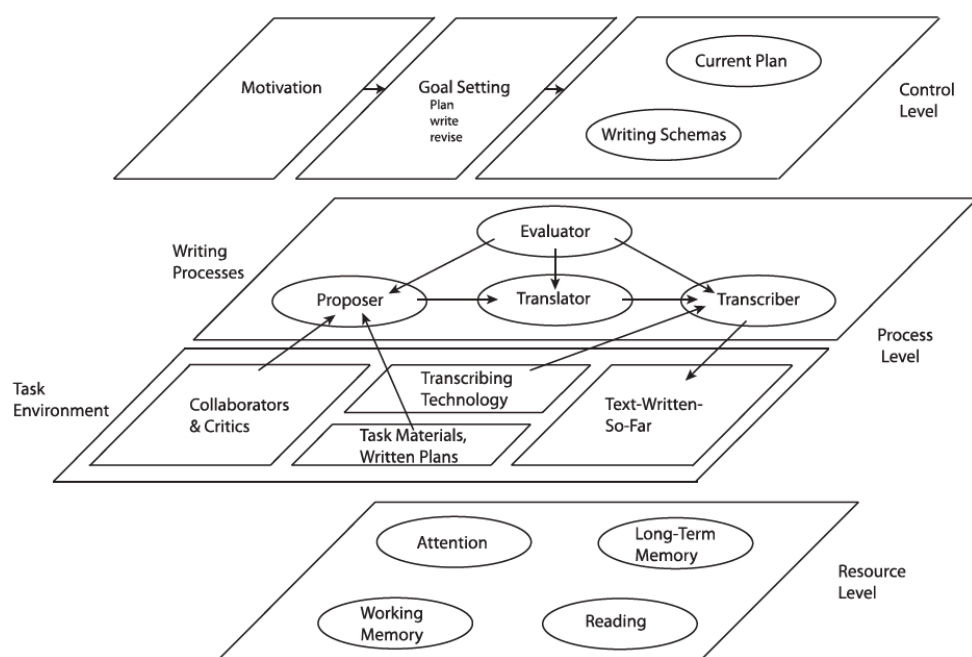
Figure 1.3. Hayes (2012) three-level cognitive process model of writing.

1.2.1.2. Local models of skilled writing: Psychomotor model. In contrast to global

models discussed previously, local models focus more on the specific elements of the writing

process (Alamargot & Chanqouy, 2001). Although the psychomotor model by van Galen

(1991) describes both higher and lower level processes, van Galen devotes most of the model

to elaborating on the handwriting processes of written production. In this regard, the

psychomotor model is a local model. The psychomotor model features seven functional

modules ordered hierarchically with the size of the unit processed decreasing from higher to

lower processes (see Figure 1.4). Temporarily, higher order modules become active first and processing cascades down the model with multiple processes being active in parallel. Each functional module is accompanied by a buffer to mediate information flow between processing units.

The higher-level cognitive aspects of the model – *activation of intentions, semantic retrieval, and syntactical construction* – were derived from a model of oral language production (Levelt, 1989) rather than written production models. The equivalent process from written production are planning and translation (Hayes and Flower, 1980). Van Galen did not specify why he followed an oral language rather than written language production architecture, but this decision seems inappropriate given that detailed models which include higher level processes were already in existence (e.g., Flower & Hayes, 1980).

After higher level processes come transcription processes, often conceptualised as lower level processes (Berninger et al., 1992). The first of these is the spelling module. Van Galen (1991) acknowledged the complexity of spelling processes in adults and, for parsimony, elected to conceptualise spelling as a singular module. Since the conception of this model, much work has been conducted to better understand spelling processes that contribute to spelling production (Rapp, Epstein, & Tainturier, 2002). Whilst different theories of spelling propose different architectures, lexical, phonological, morphological, and orthographic knowledge processes are implicated in spelling production.

Van Galen (1991) suggested that words were activated as letter strings which are then processed in three discrete motor processing modules, the main focus of this model. The first module is concerned with *allograph selection*. Each letter varies in its motoric and visual properties and allographs are the specific version of the letter. For example, $<l>, <L>, <\iota>$ are all allographs of the same letter. An abstract sensorimotor map or motor programme of each allograph containing information of its shape, stroke sequence, and direction is stored in

long term memory of experienced writers (Teulings, Thomassen, & van Galen, 1983). Accordingly, at allograph selection, the motor programmes of allographs of each grapheme in the word are activated. In a separate module, *size control*, task-specific parameters such as size and speed are coded individually for each allograph. At the final stage is the recruitment of the relevant arm, wrist, and hand muscles to produce strokes required to form the letter.

The rationale for three separate motor processing modules was based on findings from experimental and neuropsychological studies identifying dissociations between three distinct processes (Margolin & Wing, 1983; Pick Jr. & Teulings, 1983; van Galen & Teulings, 1983). Van Galen and Teulings (1983) measured the time adults took to begin writing (onset latency) when copying letters which differed in their orientation (forward and reversed), size (large and small), and slant (varying angles). The authors concluded that differences in onset latencies in each of these manipulations reflected discrete allograph (orientation), size control (sizes), and muscular adjustment (angles) stages.

Unlike Hayes and Flower's (1980) model, this model goes to some length to elaborate on the complex motor processes involved in written production using converging evidence from experimental and neuropsychological studies. However, the psychomotor model is not without its limitations. Kandel and colleagues have since challenged van Galen's view that words are activated as linear sequences of letters and argued instead that they are activated as functional linguistic units such as graphemes, morphemes, and syllables (Kandel, Hérault, Grosjacques, Lambert, & Fayol, 2009; Kandel, Peereman, Grosjacques, & Fayol, 2011; Kandel, Spinelli, Tremblay, Guerassimovitch, & Álvarez, 2012).

A second issue is the lack of a feedback mechanism in the lower level modules of the model. Van Galen (1991) himself noted "a serious limitation of the model in its present form is that it does not represent any feedback process" (p. 185). Studies of adults (patient and non-patient samples) and children have shown perceptual (visuospatial and kinaesthetic)

feedback are important for online correction during letter formation (see Danna & Velay, 2015 for review) and so it is important that these processes are accounted for. Despite these limitations, van Galen's (1991) detailed description of skilled motor process during written production remains relevant in understanding writing and more specifically handwriting in children and adults (Palmis, Danna, Velay, & Longcamp, 2017).

PROCESSING MODULE	UNIT SIZE	BUFFER STORAGE
Activation of Intentions	Ideas	Episodic Memory
Semantic Retrieval	Concepts	Verbal Lexicon
Syntactical Construction	Phrases	Short-Term Memory
Spelling	Words	Orthographic Buffer
Selection of Allographs	Graphemes	Motor Memory
Size Control	Allographs	Motor Output Buffer
Muscular Adjustment	Strokes	

Real-Time Trajectory Formation

Figure 1.4. Van Galen's (1991) hierarchical psychomotor model of writing.

Since the conception of van Galen's (1991) model, neuropsychological studies of patients with brain injury have delineated many of the spelling and motor processes related to handwriting production (Ellis, 1993; Rapp & Caramazza, 1997). Of particular interest is the connection between central spelling processes and more peripheral motor processes. Rapp

and Caramazza (1997) reported on two sets of patients with acquired dysgraphia who made a large number of letter substitution errors (replacing one letter for another). Further analyses of these patients revealed that one set had difficulties in both oral and written production and errors in these patients increased with the length of the word. Yet, despite these errors, in the main, the consonant-vowel structure of the words remained unaffected and substitutions were not related to the motoric similarity of the letters. Conversely, in the second set of patients, only written production was impaired. These patients would more frequently substitute letters with motorically similar items without regard for the consonant-vowel structure or the length of the words. This dissociation between the two groups in the nature of substitution errors led Rapp and Caramazza (1997) to suggest the former set of patients had impairments in the processing of abstract graphemic representations (e.g., identity and order) in what the authors termed the *graphemic buffer*. However, the latter set of patients were determined to have impairments in processing abstract motoric representation (e.g., number of strokes) in what the authors identify as the *allographic store* which is analogous to van Galen's (1991) allograph selection. The graphemic buffer is therefore likely to be a bridge between central spelling processes and more peripheral motor processes.

To sum up, since the conception of Hayes and Flower's (1980) model there has been considerable progress made in refining our understanding the global processes of writing. With the evolutions of these global models, two key components have become apparent. The first is that cognitive processes which are not specific to writing, such as memory and attention, make an important contribution to facilitating written production (Hayes, 1996; Hayes, 2012; Kellogg, 1996; van Galen, 1991). The second is that transcription skills are vital components of written production (Bourdin & Fayol, 1994; Hayes, 2012). These low-level skills are complex in themselves as captured in van Galen's model (1991). The

importance of adequate development of spelling and handwriting in children is apparent in models of writing development.

1.2.2. Writing Development: The Importance of Spelling and Handwriting

Writing development does not simply reflect a reduced version of skilled writing and so models of skilled writing do not adequately explain writing processes in children (Berninger & Swanson, 1994). As such, several models have attempted to model writing development. Similarly to the models of skilled writing, some models of writing development focus on global processes (e.g., Berninger & Swanson, 1994) whilst others focus on local processes (e.g., Berninger, Mizokawa, & Bragg, 1991). As will become apparent, all models highlight the importance of developing fluent and accurate spelling and handwriting.

1.2.2.1. The simple view of writing. In their study of literacy development, Juel, Griffith, and Gough (1986) were interested in the related development of reading and writing acquisition. In relation to the former skill, Gough and Tunmer's (1986) simple view of reading suggests that the two vital components of reading development are *decoding ability* and *listening comprehension*. Under this view, decoding is necessary but not sufficient for reading. In other words, for reading to be successful, children must not only be able to read the word (decoding) but understand what is being read (underpinned by listening comprehension), also. The authors also highlight that decoding and listening comprehension have complex underlying mechanisms themselves.

For writing, Juel et al. (1986) draws parallels with reading by suggesting writing requires *spelling* and *ideation*. In this model, spelling and ideation of writing are analogous to decoding and listening comprehension of reading. That is, spelling is necessary but not sufficient for writing and both spelling and ideation are driven by complex underlying mechanisms. Linking reading and writing, spelling and decoding are hypothesised to share

what the authors describe as *cipher knowledge* – composed of phoneme awareness and print exposure – and *lexical knowledge* (knowledge of which rules apply to specific words).

A combined writing and reading model was tested longitudinally in children between Grades 1 (equivalent to Year 2 in the UK) and 2 (Year 3 in the UK). In line with the predictions of the model, cipher and lexical knowledge were important determinants to both spelling and decoding ability. Furthermore, both spelling and ideation predicted writing quality. The relative weight of these skills on writing changed with age whereby spelling explained the most variance in first grade but ideation was the best predictor of writing in the second grade. The authors interpreted this in the context of a limited capacity system – similarly to Chenoweth and Hayes (2001) – and suggested lower-level spelling skills must become automatic to reduce the cognitive load and free up capacity for higher level ideation mechanisms. Despite decoding and spelling sharing common determinants, there was a weak relationship between reading and writing skills in the early grades which the authors attribute to the large differences in higher level skills between reading and writing.

The simple view of writing offers a model which is based on empirical findings and one that can be objectively falsified. Furthermore, it offers a link between reading and writing and elaborates on the mechanisms of spelling ability. However, what it brings in objectivity it lacks somewhat in specificity. At the higher level, this model does not elaborate on how ideas are generated, planned, and translated into linguistic information (c.f., Flower & Hayes, 1980). At the lower level, the model describes at length how spelling is vital for writing, but does not account for graphomotor conversion, or handwriting, which is also a significant predictor of writing quality in children at this age (Berninger et al., 1992; Graham, Berninger, Abbott, Abbott, & Whitaker, 1997; Kim & Schatschneider, 2017).

1.2.2.2. Modifying the cognitive process model. In a similar vein to Juel et al. (1986), Berninger and colleagues were committed to elucidating the determinants of writing

development (Abbott & Berninger, 1993; Berninger et al., 1992; Berninger, Cartwright,

Yates, Swanson, & Abbott, 1994). This large body of work has culminated in several models

and theories of writing. Early attempts at modelling writing development focused on adapting

Flower and Hayes (1980) cognitive process model (e.g., Berninger & Swanson, 1994).

Recognising that writing processes were different in developing than in skilled

writers, Berninger and Swanson (1994) and later Berninger, Abbott, Whitaker, Sylvester, and

Nolen (1995) proposed eight adaptations to Hayes and Flower's (1980) model to account for

developing writers. The adapted model is architecturally ordered like Flowers and Hayes'

(1980) model (see Figure 1.5), although there were some important changes. A key

elaboration made to this model is in its *translating* component.

As noted earlier, Hayes and Flowers (1980) did not provide a great deal of specificity

regarding *translation* processes, instead, assuming these processes did not influence the

skilled writer (Hayes, 2012). Based on their observations of developing writers, Berninger et

al. (1992) discriminated between several translation-related processes. Specifically, they

found some children could generate ideas but were not able to deliver these ideas in a

linguistically coherent way, suggesting discrete processes were responsible for idea

generation and text generation. Further observations revealed that some children were unable

to write legibly with invented spellings but could read their text fluently whereas others were

able to write legibly and use appropriate spelling but were unable to produce text. Based on

these findings, Berninger and Swanson (1994) made the distinction between text generation –

translating ideas into language – and transcription, converting language into symbols on the

page. Notably, this modification was not made to models of skilled writing until Chenoweth

and Hayes (2001).

In addition to providing greater specificity to translation processes, Berninger and

Swanson (1994) also proposed a framework of the developmental trajectories of different

aspects of the models. Based on their cross-sectional studies of children in Grades 1 to 9 (Years 2 to 8 in the UK; Berninger et al., 1992; Berninger et al. 1994) transcription skills are the first to emerge followed by text generation at the word, sentence, and discourse levels. The development of translation processes precedes the development of planning which is followed by revision processes. Surmising that revision processes are the last to develop is in accordance with observations made by Juel et al. (1986) who noted that young children hardly ever revise their writing.

During early phases of writing development, Berninger and Swanson (1994) found that short-term memory played a pivotal role, whereas working memory was important for higher level processes. The authors followed a limited capacity view and proposed that at this early stage of development, children's capacity is taken up by the cognitively costly non-automatized transcription skills (see also Bourdin & Fayol, 1994). When transcription skills become automatised resources are assumed freed to be allocated to higher level writing processes under the control of working memory.
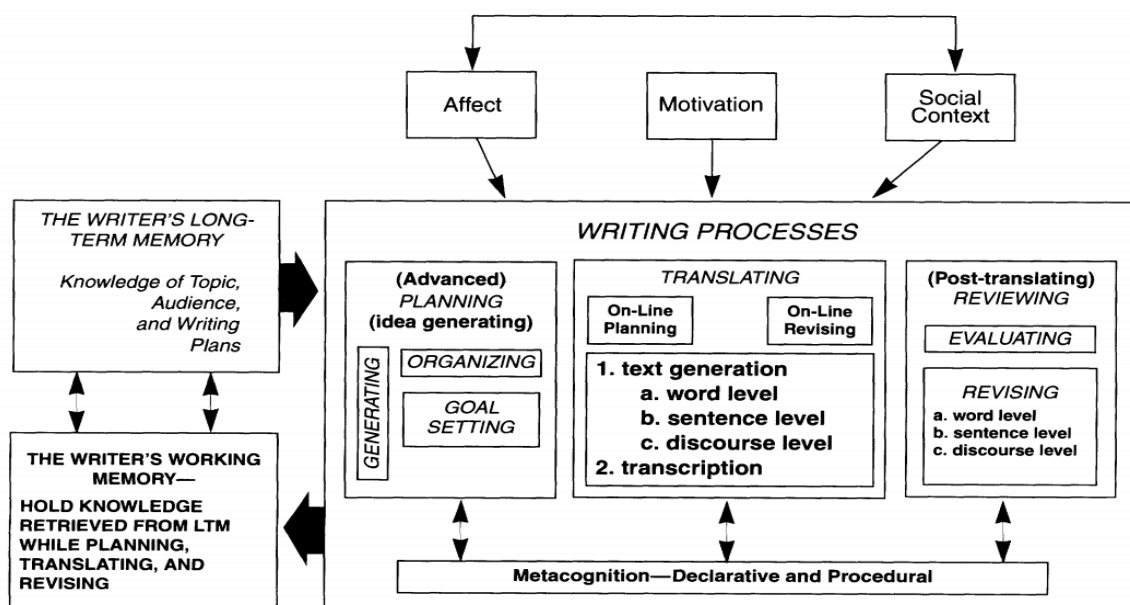
Figure 1.5. Berninger and Swanson's (1994) modified cognitive process model (Hayes & Flower, 1980) to explain developing writing.

1.2.2.3. Further simple views of writing. Berninger and Amtmann (2003) and later Berninger and Winn (2006) sought to align the simple view of writing proposed by Juel et al. (1986) with their modified version of Hayes and Flower's (1980) model to identify components of writing which could be targeted for identification and intervention in children with writing difficulties. In this tripartite model (see Figure 1.6), *transcription* and *executive functions* are foundational skills for *text generation* facilitated in a working memory environment.

The unique contribution made by transcription-related skills (e.g., orthographic knowledge and fine motor skills) to compositional quality in young writers (e.g., Berninger et al., 1995; Graham et al., 1997) led to the inclusion of transcription skills in the model. According to this model, transcription skills are responsible for translating generated ideas into functional language on the paper. Executive functions are responsible for overseeing the writing processes planning, translating, and reviewing of writing. Initially, the authors suggested that these processes are fulfilled by external actors such as teachers, but with instruction and cognitive development these functions become fulfilled internally via self-regulation (see Santangelo, Harris, & Graham, 2016). In their later, *not-so-simple* version of the model (see Figure 1.6), Berninger and Winn (2006) hypothesised that self-regulation executive function is an attention-based system that includes selective attention (focusing on what is relevant and inhibiting what is not), sustained attention (remaining on task), switching attention, and what the authors term conscious attention involving metalinguistic and metacognitive awareness.

Working memory also assumes a core role in this model as demonstrated by its central placement in the middle of the triangle. It is hypothesised to serve several functions including to activate information stored in long term memory during composition (e.g., phonological knowledge) as well as to activate short-term memory for reviewing and revising

text. Working memory is assumed to link verbal working memory with executive functions (Berninger & Amtmann, 2003; Berninger & Winn, 2006). Similarly, to other models of writing, working memory is assumed to have a limited capacity. Accordingly, in early writers, transcription skills are believed to absorb much of this limited capacity system, and automatization of these skills is necessary to free up working memory resources for the development of text generation skills.
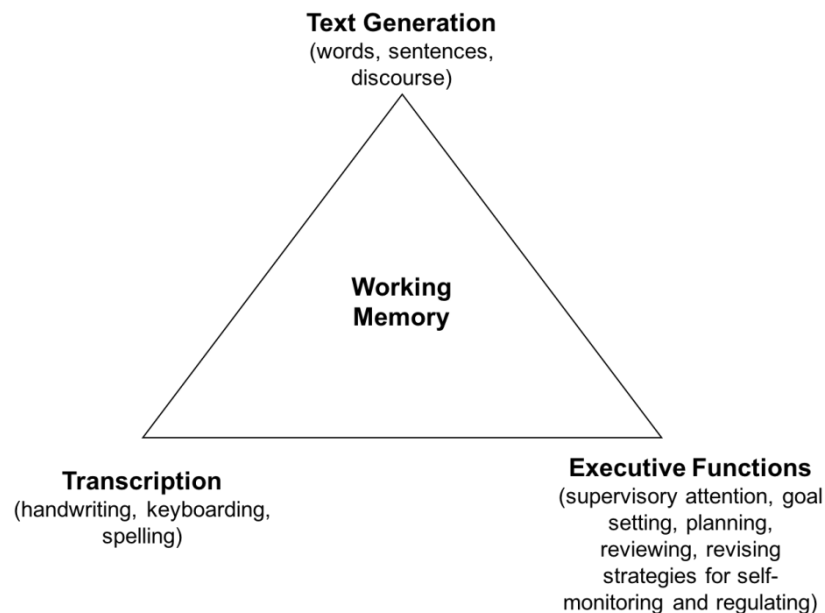
Figure 1.6. The not-so-simple view of writing by Berninger and Winn (2006).

1.2.2.4. Developing writing in a limited capacity system. Working memory plays a role in several of the models of skilled and developing writing reviewed so far (Berninger & Amtmann, 2003; Chenoweth & Hayes, 2001; Hayes, 2012; Hayes & Berninger, 2014; Swanson & Berninger, 1994; van Galen, 1991). Due to the prominence of working memory in the writing literature, it is important to properly consider the role of working memory and the assumptions that underpin this construct when explaining writing development.

As has become clear, theories of writing development typically follow a limited capacity view (e.g., McCutchen, 1996, 2000, 2011; Olive, 2014). Under this view, several assumptions about working memory and writing processes are made, specifically (a) working memory can be utilised by writing process (e.g., Berninger & Amtmann, 2003), (b) working memory is a limited capacity system (e.g., Berninger, 1999), (c) writing processes exert a cognitive load onto the working memory system, and (d) with increasing load, the available working memory resources for other processes in the system are reduced (Berninger, Abbott, Abbott, Graham, & Richards, 2002; Bourdin & Fayol, 1994; McCutchen, 2011).

As was highlighted in Berninger and Swanson (1994), in young writers, transcription processes are thought to be particularly effortful and cognitively costly placing high demand on limited working memory resources. As handwriting and spelling become automatized and children become more fluent at executing these skills, transcription processes place a smaller cost on working memory capacity, freeing up resources for higher level processes (Berninger et al., 2002; Berninger & Swanson, 1994; Bourdin & Fayol, 1996; McCutcheon, 2011).

Evidence for this position comes from studies investigating transcription processes. For example, studies of predictors of writing quality report that as children's handwriting and spelling become more fluent, these skills account for less variance in writing quality (Berninger & Swanson, 1994). A meta-analysis of writing interventions by Graham, Mckeown, Kiuhara, and Harris (2012) found eight studies where targeting transcription skills improved writing quality (average weighted effect size = .55). Furthermore, the recall of words was lower in a written condition than an oral condition in children but not in adults. Recall amongst adults did drop when they were asked to write in an unfamiliar script type using cursive capital letters (Bourdin & Fayol, 1994). Together, these findings were taken to suggest that transcription skills placed a larger load on working memory in children because adults have automatized their skills, freeing up resources. Yet, when automatization is

removed – by writing in an unfamiliar script – transcription skills place a larger cognitive load in adults, also.

The limited capacity account unifies several theories of writing development (e.g., Berninger & Swanson, 1994; Berninger & Amtmann, 2006). However, the theory is not without some limitations. A key limitation of this explanation is that it is hard to falsify. As Torrance and Galbraith (2006) note "it is sometimes difficult to imagine patterns of the data in research of this kind that could not be explained by some combination of capacity and automaticity effects" (p. 4). A related issue is the conflation between working memory and attentional processes. Specifically, working memory is assumed by some to coordinate between writing processes, presumably as part of the central executive (Olive, 2014). However, others have suggested that the coordination is completed by attentional processes (e.g., Hayes & Berninger, 2014; Berninger & Winn, 2006). It is therefore unclear whether either attentional, working memory, or attentional and working memory processes coordinate writing processes.

Alternate explanations to a limited capacity view include *interference* or *cross-talk* account which provide an equally good fit to the findings (Torrance & Galbraith, 2006). From an interference perspective, the output of one process may interfere with another process. An example in the case of transcription skills may be that online resolution of spelling processes interferes with the graphomotor expression of the word. Such findings have been recently reported in psycholinguistic studies examining the temporal bases of spelling execution in adults and children (e.g., Kandel & Perret, 2015; Roux, McKeeff, Grosjacques, Afonso, & Kandel, 2013).

1.2.2.5. Summary. The evolution of models of skilled writing demonstrated the importance of transcription skills in writing. This point is captured to a greater extent in models of writing development where transcription skills are a key component of

development (Amtmann & Berninger, 2003; Berninger & Swanson, 1994; Juel et al., 1986).

The automatisation of spelling and handwriting skills appears to be critical for the

development of higher-level writing skills (Berninger, 1999; McCutcheon, 2011). Spelling

and handwriting are separable but highly related skills with complex mechanisms (e.g.,

Berninger et al., 1992; Juel et al., 1986), which are not described sufficiently in models of

writing development. Thus, in the next section I will discuss the typical development of

spelling and handwriting skills.

1.3. Typical Spelling and Handwriting Development

As was apparent in the previous analyses of writing models, spelling and handwriting

are important determinants of writing ability. Spelling and handwriting skills hold a special

position as being the foundation on which higher level skills are built (Berninger &

Amtmann, 2003). Thus, typical development of these separate but highly related constructs is

important for the development of higher-level writing processes. Although children learn to

spell and handwrite at the same time, the development of these skills has been charted

separately. Psycholinguists and educationalists have focused on spelling development

whereas psychomotor specialists have primarily been interested in mapping handwriting

development. In this section, I outline some of the relevant aspects and theories of typical

spelling and handwriting development.

1.3.1. Spelling Development

Proficient spelling requires children to build lexical, orthographic, phonologic, and

morphologic, and syntactic knowledge of written words (Bahr, Silliman, Berninger, & Dow,

2012; Bourassa & Treiman, 2003). The growth of spelling skills is moderated by the

complexity of the system the child must learn. The complexity of the system is highlighted in

the *consistency* between sound (phoneme) and letter (grapheme) mappings (Caravolas, 2004).

In more consistent languages such as Welsh the mappings between phoneme-to-grapheme is close to one-to-one. That is, one sound is mapped to one letter. In more inconsistent languages such as English there are multiple mappings between phoneme-and-graphemes whereby sounds are mapped to multiple letters (although the sound-letter consistency improves in the context of larger orthographic units; Kessler & Treiman, 2003). This means that children's literacy develops faster in consistent than inconsistent orthographies. Nevertheless, becoming a fluent and accurate speller takes years to master (Treiman, 2017a).

Theories explaining how we master this complex skill take several forms including (a) stage theories (e.g., Ehri, 1997), (b) connectionist theories (e.g., Houghton & Zorzi, 2003), (c) dual-route theories (e.g., Sprenger-Charolles, Siegel, & Bonnet, 1998), (d) triple foundation model (Caravolas & Samara, 2015), and (e) integration of multiple patterns (Treiman & Kessler, 2014). In what follows, I elaborate more on the latter three theories. These three theories are principally concerned with spelling development in different ways. Whilst dual-route theories are concerned with processes involved in spelling production the triple foundation and the integration of multiple patterns is less concerned with production of spelling and more concerned with the skills necessary for early literacy development.

1.3.1.1. Dual-route theory. The dual-route account of spelling development follows the basic premise of dual-route models of skilled reading and spelling (e.g., Coltheart, 2005; Rapp et al., 2002). These theories attempt to explain the process by which we retrieve or construct the word to be spelt. According to dual route theorists, spelling follows one of two interactive routes, the *lexical* and *sub-lexical route*. Spellings of words which the individual is familiar with, regardless of their complexity (e.g. sound-letter consistency) are encoded into memory stores, or, *lexicons*. Phonological information of the word is stored in the *phonological lexicon* whilst the orthographic information is stored in the *orthographic lexicon.* Accordingly, when spelling the familiar word, the phonological lexicon is activated,

in turn this activates the orthographic lexicon and the spelling is retrieved and spelt correctly.

However, if the word is new to the individual or the representations are incomplete (e.g., they

are familiar with hearing the word but not writing it) then the second, sub-lexical, route is

used. The sub-lexical route does not rely on prior knowledge of the word, but instead builds

the word by applying common phoneme-to-grapheme rules. This means that unfamiliar

consistent words would be spelt correctly whereas unfamiliar inconsistent words could be

spelt incorrectly with phonologically plausible errors (Rapp et al., 2002).

Developmental versions of this model propose young children spell initially using the

sub-lexical route, as they have not yet acquired orthographic knowledge of the word. In as

little as three months of instruction, however, children begin to develop phonological and

orthographic knowledge and begin to use the lexical route (Martinet, Valdois, & Fayol, 2004;

Sprenger-Charolles, Siegel, Béchennec, & Serniclaes, 2003; Sprenger-Charolles et al., 1998).

Evidence for this position is that children's spelling accuracy is sensitive to the word

frequency early in instruction and this frequency effect increases with age (Martinet et al.,

2004). To enable the use of the sub-lexical route, the theory also assumes that children must

have some letter knowledge and phoneme awareness.

However, in these models, the consistency of a word is often considered dichotomous

and there has been little elaboration of how the sublexical route treats varying consistencies.

Furthermore, these models do not explain how children acquire other types of knowledge

(e.g., morphology). The lack of clarity in this regard makes it hard to generate predictions

about how spellings are learned in the model (Treiman & Kessler, 2014).

1.3.1.2. Triple foundation model. The triple foundation model differs from the dual-

route model in that it is less concerned with describing the processes by which spellings are

produced and is more concerned with the component skills that contribute to early literacy

development across languages. The triple foundation model proposes three cognitive

processes – *letter knowledge*, *phoneme awareness*, and *rapid automatized naming (RAN)* – underpin early reading and spelling development across all languages (Caravolas & Samara, 2015). The architecture of the model is based on a strong bank of empirical studies seeking predictors of literacy development (e.g., Caravolas et al., 2012). Both children's knowledge of alphabet letter's names and sounds (letter knowledge) and their ability to manipulate spoken sounds (phoneme awareness) individually predict development of spelling skills at the start of formal literacy instruction (Byrne, 1998; Caravolas, Hulme, & Snowling, 2001). A third cognitive determinant of literacy, RAN also predicts the growth of spelling ability (Georgiou, Torppa, Manolitsis, Lyytinen, & Parrila, 2012). There is some debate as to what processes RAN captures (Decker, Roberts, & Englund, 2013), but growing evidence suggests rapid naming taps the rapid cross-modal mapping between phonological and visual (orthographic) information (Lervåg & Hulme, 2009; Vaessen, Gerretsen, & Blomert, 2009).

Cross linguistic studies have demonstrated the universality of the three foundation skills in children's spelling development. Caravolas et al. (2012) examined the predictive nature of children's letter knowledge, phoneme awareness, and RAN skills – along with their existing spelling knowledge – when learning alphabetic orthographies differing in consistency (English, Spanish, Czech, and Slovak) before beginning formal instruction. Together, the model predicted 63% of the variance in spelling 10 months later (Caravolas et al., 2012). Similarly, Moll et al. (2014) also found phoneme awareness and RAN to predict spelling development in older children (letter knowledge was not included as children typically reach ceiling on this measure early in literacy development). However, direct testing of the model in non-alphabetic orthographies is needed. Nevertheless, the triple foundation model presents a strong evidence-based case that early spelling in children utilises letter knowledge, phonological, and RAN skills.

1.3.1.3. Integration of multiple patterns (IMP). Treiman and Kessler (2014) take a different view to that of dual-route theorists and assert that there is more consistency in the writing system than previously thought (Kessler & Treiman, 2003). Furthermore, like the triple foundation model, the IMP is not concerned with how the spelling processes develop but takes a holistic view of how knowledge is acquired for spelling.

The central tenet of this model is that children learn multiple patterns that are applied in some (probabilistic) or all contexts (deterministic). Children learn about these patterns implicitly via statistical learning or explicitly via instruction. The probabilistic nature of this learning means children learn best when converging patterns support the use of a specific pattern. Where there is conflict or lack of knowledge (e.g., an unfamiliar word with an inconsistent phoneme-grapheme mapping in it), children will use the pattern they are most familiar with in that context (Treiman, Decker, Kessler, & Pollo, 2015). That is, they will use the spelling pattern with the highest probabilistic value.

Treiman and Kessler (2014) propose that children learn two categories of patterns. The first category is concerned with learning the *outer form of writing* which describes the graphic patterns of writing. Graphic patterns of writing include letter shapes and plausible letter sequences (graphotactics). In relation to the shapes of letters, one pattern is the orientation of the letter. Most letters in the English alphabet are right-facing. It follows that young children are more accurate at forming right-facing letters than left facing (Fischer, 2013; Treiman & Kessler, 2011). This phenomenon can be explained using an IMP account, where greater accuracy for right-facing than left facing letters suggests children have implicitly learned the pattern that letters are more likely to be right facing (Treiman and Kessler, 2011).

According to the IMP, children begin to learn about the outer forms of writing early in development. Despite being unable to produce recognisable letter forms, children aged

between 2 and 4 years old are able produce marks which are distinct from drawing (Treiman & Yin, 2011) as well as produce marks in the left-to-right direction (Rowe & Wilson, 2015). Furthermore, children at this age will often include letters and letter clusters that are frequent in their language (Kessler, Pollo, Treiman, & Cardoso-Martins, 2013). Collectively these findings suggest children learn some basic visuomotoric and graphotactic patterns of letters prior to learning the relationship between symbols and language (i.e., alphabetic principle).

The second category of patterns children learn is concerned with the *inner form of writing* which describes the connections between written characters and language (Treiman, 2017b; Treiman & Kessler, 2014). Here too, the IMP predicts that children will learn the patterns of letters and sounds probabilistically. This would mean children are more likely to use letters in a phonologically appropriate way if they are familiar with them. Indeed, early spellers who have not yet grasped all phoneme-grapheme mappings are more likely to use a letter in a phonologically appropriate manner if it was the initial letter of their first name (Both-de Vries & Bus, 2008), suggesting children are sensitive to patterns of phonemes. Indeed, with experience, children also become sensitive to orthographic and morphological patterns (Kessler, 2009; Treiman, 2017b). In this manner, the IMP provides a more holistic explanation of spelling development than dual-route theories by accounting for the development of aspects of spelling such as graphotactic and morphological knowledge (Treiman, 2017a).

The three models of spelling development reviewed here conceptualise how we acquire complex spelling knowledge differently. The first, dual-route theory, provided a theoretical account of how spelling processes develop and operate in children. The second, triple foundation model, and third, IMP, are more concerned with how we develop knowledge necessary for spelling. The triple foundation model emphasised the importance of the foundational skills underpinning spelling (and reading) development, namely letter

knowledge, phonological skills, and rapid naming. The IMP account was less concerned with foundational skills or the process of spelling but suggested we learn and integrate patterns of language to spell. A unifying aspect of the three accounts is the emphasis placed on phonological and letter-sound knowledge for successful growth of spelling.

1.3.2. Handwriting Development

Handwriting is a complex skill which takes many years to learn (Graham, Berninger, Weintraub, & Schafer, 1998; van Galen, 1991). It is conceptualised in terms of legibility (readability) and fluency (speed). This thesis is concerned with the development of typical and atypical handwriting legibility and fluency. When evaluating studies of handwriting development, it is important to consider the influence of highly variable instructional practices and methods for measuring handwriting. Therefore, it is prudent to outline briefly variations in instructional practices and methods for measuring handwriting before focusing on the growth of handwriting.

1.3.2.1. Handwriting instructional practices. Handwriting skills benefit from regular and evidence-based instruction (Graham & Harris, 2005; Jones & Christensen, 1999; Vander Hart, Fitzpatrick, & Cortesa, 2010). Furthermore, there is some evidence to suggest that teacher's attitudes and competence also influence handwriting ability (Graham et al., 2008). A large survey of American primary school teachers ($N = 169$) revealed that although 90% of respondents taught handwriting, there was considerable variability in the frequency, duration, and method of instruction. Moreover, only 12% of teachers felt their teacher training had equipped them suitably to teach handwriting. Such variability in the quantity and quality of instruction will likely lead to variability in handwriting abilities in school aged children.

In Wales, teachers are expected to teach to the national curriculum set by the Welsh Government. The handwriting specific benchmarks per school year group included within

this curriculum and are outlined in Table 1.1. To my knowledge, there is currently no empirical research examining the frequency, duration, or method of instruction of this handwriting curriculum in Wales. However, a small-scale survey of primary teachers in England ($N = 39$) revealed similar findings to Graham et al. (2008). The survey of teachers in England revealed there to be large variation in school handwriting teaching policies, not much time was spent on handwriting in the classroom, and teachers did not feel well prepared for teaching handwriting (Stainthorp, Barnett, Henderson, & Scheib, 2006). It is highly probable that these findings are applicable in Wales and are likely to effect handwriting development and possibly lead to considerable variation in handwriting abilities. Further work should investigate the role of instructional practices in handwriting development.

Table 1.1.

Learning Wales (2014) National Curriculum Handwriting Benchmarks

	Attainment Criteria
Nursery	• Use a pincer grip to hold writing tools appropriately.
Reception	• Hold writing tool appropriately. • Write from left to right.
Year 1	• Appropriately form upper- and lower-case letters with clear shape and correct orientation.
Year 2	• Accurately form upper- and lower-case letters. • Letters should be a consistent size.
Year 3	• Produce legible handwriting. • Appropriately join letters in some words.
Year 4	• Produce legible handwriting. • Handwriting may be cursive.
Year 5	• Produce legible handwriting. • Handwriting may be cursive. • Handwriting should be increasing fluent.

| Year 6 | • Produce legible handwriting.
• Produce fluent handwriting. |

1.3.2.2. Measuring handwriting legibility and fluency. There is also considerable variation in how teachers and researchers assess handwriting (Graham et al., 2008). Handwriting legibility is typically measured using a rating scale applied to text. Several rating scales have been developed for this purpose and are categorised as either *global* or *analytic scales.* Global scales garner an overall impression of how readable the production is whereas analytic scales examine constituent parts of handwriting that contribute to the readability. Poor ease of use, low/unreported reliability and validity, and little/no evidence of standardisation in most current handwriting legibility scales (Rosenblum, Weiss, & Parush, 2003) have meant that researchers have focused predominantly on measuring handwriting fluency (Abbott & Berninger, 1993).

Handwriting fluency is easier to objectively quantify. Fluency is typically assessed using simple writing tasks (e.g., copying) under timed conditions where the main outcome measure is to count the number of letters/words produced within the duration of the task (Abbott & Berninger, 1993; Barnett, Henderson, Scheib, & Shulz, 2007). In addition to traditional measures of fluency, technological advances mean that handwriting fluency can also be measured in real time using digisting tablets and pen tracking. Pen tracking also involves the individual completing simple writing tasks (e.g., copying). Software such as Eye and Pen (Alamargot, Chesnet, Dansac, & Ros, 2006) records the coordinates of the pen travelling on the tablet and extracts several parameters. Common fluency related parameters in the literature include pauses (Prunty, Barnett, Wilmut, & Plumb, 2013; Sumner, Connelly, & Barnett, 2013, 2014), duration (Kandel, Lassus-sangosse, Grosjacques, & Perret, 2017), speed (Prunty et al., 2013; Sumner et al., 2013; 2014), and fluency (Kandel & Perret, 2015) of pen movements. These parameters are often referred to as *process* or *online measures* in

the sense that they are capturing processing as it is occurring whereas traditional fluency measures are referred to as *product* or *offline measures*.

1.3.2.3. Growth of handwriting legibility and fluency. Few studies have charted the growth in handwriting legibility and fluency and, to my knowledge, no studies have examined handwriting development longitudinally. However, a large cross-sectional study by Graham et al. (1998) measured handwriting legibility and fluency of 100 American children in each school year group between Grades 1 to 9 (5 to 14 years old) and provides a good description of handwriting growth.

Graham et al. (1998) applied a global handwriting legibility scale separately to copying, writing to dictation, and composition tasks and found little growth in handwriting legibility between the ages of 5 to 7 years old. After the age of 7, legibility improved with every school year until the age of 11 when it plateaued (Graham et al., 1998). These findings contrast with other studies which have reported larger increases in legibility during primary school years (e.g., Hamstra-Bletz & Blote, 1991). However, the differences between studies are most likely due to the type of handwriting legibility scale used. Graham et al. (1998) used a global handwriting scale whereas analytic scales used in other studies are more likely to be sensitive to subtle growth.

Handwriting fluency was assessed by Graham et al. (1998) by calculating the number of letters formed correctly in 90 seconds. Using this product fluency measure, the growth in handwriting fluency was found to be non-linear. Between the ages of 5 and 8, fluency increased consistently between the grades. The rate at which fluency increased per year slowed between 8 and 10 years old. After the age of 11, the rate of fluency increased again until around the age of 13 when it slowed again. The plateau in performance between the ages of 8 and 11 years old is a common phenomenon and has been reported across multiple measures of fluency (Meulenbroek & van Galen, 1990; Thibon, Gerber, & Kandel, 2018).

Based on Graham et al. (1998) findings that the legibility and fluency grow at different rates, it appears that legibility and fluency are separable skills with different patterns of development. This view is further promoted by the weak correlation between legibility and fluency reported by Graham et al. (1998). The growth rates of both legibility and fluency also show that handwriting development is a prolonged process. This is in agreement with other studies reporting that children's handwriting does not become adult like until after 12 years of age (Thibon et al., 2018) with some suggesting complete automatisation is not achieved until the age of 15 (Accardo, Genna, & Borean, 2013). An interesting period for handwriting fluency development, though, appears to be in late primary school between the ages of eight and eleven when fluency temporarily plateaus.

1.3.2.4. Theories of handwriting development. Unlike spelling, there have been few theoretical explanations for the growth of handwriting described in the previous section. This is perhaps in part due to the lack of data of handwriting growth and in part due to the lack of objective and sensitive measures of legibility. The development of handwriting is commonly considered within a motor learning framework, but handwriting development has also conceptualised within stage theory (Berninger et al., 2006). Both the general motor framework and stage theory will be considered here. However, I believe that the IMP discussed earlier can also account for some aspects of handwriting development and so this will be discussed, also.

1.3.2.4.1. Motor learning framework. The general motor learning framework has not been conceptualised as a unitary theory of handwriting development, but it is often applied in the literature to explain the trajectory of handwriting growth (e.g., Palmis et al., 2017). It is not clear whether this theory should explain the development of both legibility and fluency. However, it most closely fits the growth of handwriting fluency presented by Graham et al. (1998).

The general motor learning framework chronicles the progression from explicit control of new motor action to the automatic control of skilled action. In building handwriting into this framework, the development of handwriting-specific motor action begins with explicit handwriting instruction, most likely to be in school. Early in development, children form letters using a ballistic or open-loop strategy where feedback is only available when the movement has completed. Initially, letter formation is marked by a stroke-by-stroke strategy that is difficult to complete accurately. Then, at approximately eight years old, children switch to *closed-loop control* and begin to make use of perceptual (visual and kinaesthetic) feedback to help guide formation. As children continue to practise, they build up motor programmes of the letters. When motor programmes begin to stabilise at approximately ten- to twelve-years-old, children rely on perceptual feedback to a lesser extent and instead use a feedforward strategy where motor programmes generate letter production automatically and accurately (Halsband & Lange, 2006; Palmis et al., 2017; Thibon et al., 2018).

The switch in strategy from a ballistic strategy to slower visual and kinaesthetic feedback accounts for the discontinuity in handwriting fluency commonly reported between ages 8 to 11 years old (e.g., Graham et al., 1998) and so may explain the non-linear trajectory of handwriting development. It provides a good explanation for the development of letter formation fluency. However, the framework does not specify how the separable construct of legibility develops nor does it explain handwriting development beyond letter formation, including handwriting of multiple letters to form words. On a related note, the motor framework ignores the co-development of spelling processes. Indeed, some authors argue that handwriting is a linguistic as well as a motor task (Berninger et al., 1992; Berninger, Abbott, Thomson, & Raskind, 2001; Kandel & Perret, 2015) and so a theory of handwriting development should also consider the growth in spelling processes, also.

1.3.2.4.2. Stage theory. In their stage theory, Berninger and colleagues focused less

on motor control aspects and more on the language aspects of handwriting in their stage

model (Berninger, 2006). Notably, in this model, development begins much earlier than at the

onset of instruction as described by the motor control framework. Handwriting development

begins as infants explore pen use via scribbling. This is followed by the toddler copying

isolated strokes in different directions. Before the age of 5 years old, Berninger et al. (2006)

describe how children learn the names of alphabet letters and fine motor control of the wrist

and fingers via simple drawing activities. At approximately seven years old, children begin to

copy letter forms legibly, can accurately write lower- and upper-case letter forms to dictation

and can accurately write the alphabet in sequence from memory.

This stage theory of handwriting development acknowledges that children begin to

understand the differences between letters and other aspects of drawing from an earlier age

(Treiman & Yin, 2011). It also places a larger emphasis on linguistic aspects of handwriting,

including learning letter-sound associations, a foundational skill for literacy development

(Caravolas et al., 2012). However, this model fails to explain the trajectory of handwriting

fluency and legibility described by Graham et al. (1998).

From the models reviewed so far, a pure motor learning explanation of handwriting

development does not account for co-development of language and literacy skills whereas the

stage theory by Berninger et al. (2006) lacks the ability to explain the non-linear growth in

handwriting fluency and legibility. An alternative way of accounting for handwriting

development is to consider handwriting development in relation to the outer form of writing

from the integration of multiple patterns (IMP) account.

1.3.2.4.3. Integration of Multiple Patterns (IMP) account. Whilst the IMP has never

been explicitly discussed in relation to handwriting development, the outer form of the

writing refers to children learning the visuomotor patterns of writing. Specifically, there are

many patterns in the graphic forms of letters. Letters share stroke patterns including ascenders, descenders, curves (arcades and garlands), and loops (Meulenbroek & van Galen, 1986) and – as discussed earlier – also share patterns of visual orientation (Treiman and Kessler, 2011). The graphic patterns of stroke and visual orientation and implicit learning of these patterns could either help develop motor programmes of the letters or support the child in making correct responses in the absence of a consolidated motor programme. Tangible evidence in support of this view, is that early writers are more likely to reverse letters with a left orientation than a right orientation (Treiman & Kessler, 2011), which suggests children are sensitive to the probabilistic pattern of a letter's orientation prior to the stabilisation of motor programmes. Clearly, the IMP does not account fully for handwriting development, but the theory offers a bridge between handwriting and spelling. Moreover, it provides testable predictions to consider handwriting development. However, to date no theory of handwriting development has considered the effects of variations in instructional practices on trajectories of handwriting development or explicitly described how these trajectories might differ between legibility and fluency.

1.4. Conclusion

This review of the literature on writing development has focused on how global and local models of skilled writing have evolved to consider the role of transcription (spelling and handwriting) skills in written production (Hayes, 2012). In turn, skilled models have influenced the development of models of writing development (Berninger & Swanson, 1994). Models of writing development emphasise that transcription skills are critical for the development of higher-level writing skills (Berninger & Winn, 2006; McCutcheon, 2011). However, models of writing development do not describe sufficiently the development of spelling and handwriting skills. Spelling and handwriting are separable but highly related complex skills that take many years to develop (Graham et al., 1998; Palmis et al., 2017;

Treiman & Kessler, 2014). In some circumstances, spelling and/or handwriting skills do not develop appropriately. There can be several reasons for atypical spelling and handwriting development including poor instruction, social economic factors, and the presence of neurodevelopmental disorders. The literature describing the effects of dyslexia and developmental coordination disorder (DCD) on the development of transcription skills is reviewed in Chapter 2.

Chapter 2

General Introduction Part 2: Neurodevelopmental Disorders and their Association with Transcription Impairments

2.1. Introduction

Chapter 1 highlighted the importance of transcription (spelling and handwriting) skills in writing development. It was also apparent that spelling and handwriting skills are complex, and the development of these skills is long. In some cases, children have difficulties in acquiring spelling and/or handwriting skills. There can be several reasons why children have difficulties in acquiring spelling and handwriting skills including poor instruction, social economic factors, and the presence of neurodevelopmental disorders. Little is known about the development of both spelling and handwriting skills amongst children with neurodevelopmental disorders.

The focus of this thesis is understanding the nature of spelling and handwriting impairments reported in two neurodevelopmental disorders, dyslexia and developmental coordination disorder (DCD; Berninger, Nielsen, Abbott, Wijsman, & Raskind, 2008; Prunty et al., 2013). As such, in this review I examine the literature describing theories dyslexia and DCD. I then build on the literature discussed in Chapter 1 by evaluating studies discussing spelling and handwriting impairments in these disorders.

2.2. Theories of Dyslexia and Developmental Coordination Disorder (DCD)

The term, neurodevelopmental disorders, is an umbrella category for several disorders of development. There have been several different classifications of what is and what is not a neurodevelopmental disorder. Some have taken the view that neurodevelopmental disorders should include disorders with a clear genetic aetiology (e.g., Prada-Willi syndrome) and those that can be explained on a wholly medical basis (e.g., cerebral palsy). Others have taken the view that neurodevelopmental disorders should be classified separately to those with a clear genetic or medical aetiology (see Thapar and Rutter, 2015). Here, I take the latter view and define a neurodevelopmental disorder as a behaviourally defined disorder whose exact

aetiology is unclear but is strongly associated with genetic/biological factors. Furthermore, impairments should occur early in development and continue into adulthood.

Several disorders fall under this definition including dyslexia, DCD, and attention deficit hyperactivity disorder (ADHD). These disorders share many commonalities, which include a high degree heterogeneity. Moreover, these impairments are often continuous rather than all or nothing and the threshold for meeting some diagnostic criteria is often arbitrarily set (Hulme & Snowling, 2009). Another commonality is that there are similar rates of occurrence in the population, with dyslexia affecting 3 - 10% (Snowling, 2013), DCD affecting 4.9% (Lingam, Hunt, Golding, Jongmans, & Emond, 2009), and ADHD affecting 5.3% (Polanczyk, De Lima, Horta, Biederman, & Rohde, 2007) of the population. The disorders are also frequently comorbid with one another (Kaplan, Wilson, Dewey, & Crawford, 1998).

Prior to detailing the current state of affairs with regards to spelling and handwriting impairments in dyslexia and DCD, it is important to elaborate on the background of dyslexia and DCD. In discussing these backgrounds, I present a very brief overview of competing theories that hypothesise about the causes of dyslexia and/or DCD. Causal theories of neurodevelopmental disorders take two forms. The first are single deficit theories which postulate a single deficit is responsible for the behavioural symptoms of the disorder. The second, multifactorial account, suggests multiple protective and risk factors act in a probabilistic manner to lead to the behavioural symptoms of the disorder. Of course, singular deficit and multifactorial accounts are not mutually exclusive, and a singular deficit can be considered within the multifactorial framework.

Historically, researchers have pursued single deficit accounts. As well as critically considering the quality of the evidence for these single deficit accounts, I consider some facets that theories of neurodevelopmental disorders should explain. That is, theories should

(a) have the capacity to explain typical as well as atypical development, (b) demonstrate that putative causes should be related to impairments forward in time, and (c) have the ability to explain most of the behavioural impairments of the disorder (see Hulme & Snowling, 2009).

2.2.1. Dyslexia

Dyslexia is a disorder primarily defined as affecting accurate and fluent word reading and spelling ability (Rose, 2009). As noted earlier, it is a common disorder which continues across the lifespan. There has been much interest in the causes of dyslexia and many theories have proposed core deficits in language (e.g. phonological), sensory processes (visual attention, magnocellular), and learning (automaticity/cerebellar). For economy, I briefly discuss only a few of these theories, phonological, visual attention, and automaticity deficit hypotheses.

2.2.1.1. Phonological deficit. Converging evidence suggests that phonological processing impairments are a core deficit of dyslexia (Vellutino, Fletcher, Snowling, & Scanlon, 2004). As discussed earlier, phonological skills, namely phoneme awareness, are a critical determinant of reading and spelling acquisition (Caravolas et al., 2012; Hulme, Bowyer-Crane, Carroll, Duff, & Snowling, 2012; Melby-Lervåg, Lyster, & Hulme, 2012). It follows then a deficit in phonological processing would impair the growth of reading and spelling skills, leading to the child meeting diagnostic criteria for dyslexia. There have been several suggestions of underlying components of phonological processing that are impaired in dyslexia (e.g., deficits in the retrieval or access of phonological representations; see Ramus & Szenkovits, 2008). Here, I take a holistic view and consider deficits in phonological processing more generally where converging evidence from multiple methods (case-control, correlation, and training) demonstrate a causal link between phonological deficits and dyslexia.

Case-control studies have reported that children with dyslexia perform significantly less well than typically developing children on many tests that require phonological processing (Fletcher et al., 1994; Ramus, Pidgeon, & Frith, 2003; Snowling, 2008; Vellutino, Scanlon, & Spearing, 1995). For example, 77% of a sample of children with dyslexia scored more than one standard deviation below the control mean on a measure of phonological processing. Case-control studies such as these demonstrate that most children with dyslexia have deficits in phonological processing, but they do not establish the cause of the deficit.

A better design for establishing causation are those employing longitudinal paradigms where performance on a measure of a deficit skill predicts impairments later in development. Accordingly, among children with dyslexia, phonological processing skills at kindergarten (Reception in the UK) explained a large amount of variance in reading scores at Grade 3 (Year 4 in the UK) and Grade 6 (Year 7 in the UK; Dandache, Wouters, & Ghesquière, 2014). Similarly, phonological skills were found to be a strong concurrent predictor of dyslexia across languages (Caravolas, Volín, & Hulme, 2005; Landerl et al., 2013).

The evidence presented using case control and longitudinal-correlational methods suggests a strong association between deficits in phonological awareness and reading and spelling. A more powerful design to infer a causal relationship between phonological processing deficits and reading impairments associated with dyslexia is using a training paradigm. Training paradigms work on the premise that causation is demonstrated if training to improve one skill also improves another. Indeed, several training skills targeting the growth of phonological processing skill have led to improvements in reading and spelling ability (e.g., Hulme et al., 2012; Wolff, 2016).

A phonological processing deficit can explain typical as well as atypical reading and spelling development. Evidence shows that phonological deficits present prior to the onset of explicit reading and spelling instruction are related to later reading and spelling abilities in

dyslexics, and training phonological processing improves literacy skills in dyslexics. Thus, the evidence suggests a causal link between phonological processing deficits and dyslexia.

Whilst most children with dyslexia have phonological deficits, not all do. In Ramus et al.'s (2003) sample, 33% of children with dyslexia did not meet the criteria for having a phonological deficit. This suggests that whilst phonological deficits appear to be a core deficit of dyslexia, there must be other causal mechanisms, also.

2.2.1.2. **Visual attention span.** As well as deficits in phonological processing, visual attention problems have been reported for individuals with dyslexia. Despite no obvious ophthalmic issues, those with dyslexia may report letters/words blurring or moving around (Stein, 2018). Several theories have proposed that visual processing deficits are causally related to dyslexia (Bosse, Tainturier, & Valdois, 2007; Stein, 2001). Here, I focus on the visual attention deficit hypothesis proposed by Bosse et al. (2007).

Visual attention span refers to the quantity of information that can be processed together in the *attentional window* (Goswami, 2015a). According to the multiple-trace memory model of polysyllabic word reading (Ans & Carbonnel, 1998) skilled reading involves a global and analytic procedure. The global procedure processes words as a whole unit and requires the attentional window to be large enough to process the whole word. If the attentional window is not large enough, the analytic procedure must process the word serially using smaller orthographic sizes (letter, cluster, syllable). These units then successively activate their phonological representation (van den Boer, van Bergen, & de Jong, 2015). Thus, it appears that the visual attention span is important in word reading.

The visual attention span is measured by briefly presenting a string of five characters (often letters/numbers) and asking participants to report either the entire string or a cued character (Bosse et al., 2007). Using this task, Bosse et al. (2007) found French and British children with dyslexia had a significantly smaller visual attention span than typically

developing children at the group level. Within the dyslexic samples, 44% and 35% of French and British children had a visual attention span, but not a phonological, deficit. Moreover, children's visual attention span explained a significant amount of variance in word reading over and above phonological processing. The authors interpreted these findings as suggesting that individuals with a reduced visual attention span must use an analytic procedure and cannot process words efficiently as whole units, resulting in atypical reading development. Thus, Bosse et al. (2007) proposed deficits in the visual attention span are causally related to dyslexia independently of phonological deficits.

Since the visual attention deficit of dyslexia was proposed, much work has established that children's visual attention span is related to their literacy abilities. Children's visual attention span has been found to uniquely predict word reading accuracy in children who were 6, 8, and 10 years old, even after accounting for the contribution phonological skills make (Bosse & Valdois, 2009). Similarly, the visual attention span also uniquely predicts word spelling in children who were 8 and 11 years old, also after controlling for phonological skills. Although, it is likely that the influence of visual attention is mediated by reading skills via the building of orthographic to phonological representations (van den Boer et al., 2015).

In a recent longitudinal study, van den Boer and de Jong (2018) examined the longitudinal relationship between children's visual attention span and reading ability between the ages of 8 and 10 years old. Using a cross-lagged panel design, visual attention span was found to significantly predict unique variance in word reading concurrently, however, when controlling for autoregressive effects of reading, visual attention span at 8 years old did not predict word reading at 10 years old. This finding raises questions over the causal relationship between visual attention span and later reading ability and therefore between visual attention span and dyslexia. Goswami (2015) questioned the causal role of visual attention span in dyslexia and suggested visual attention span reflects lower reading

experience rather than being causal of atypical reading development (Goswami, 2015b; see also Hawelka & Wimmer, 2008; Lobier & Valdois, 2015).

Despite questions about its causal role in reading performance, the visual attention span deficit hypothesis has been associated with typical and atypical literacy development. Furthermore, visual attention span deficits have been found in a large minority of children with dyslexia. However, there is a paucity of longitudinal and training data to provide conclusive evidence of a causal link between visual attention deficit and dyslexia. Therefore, the visual attention deficit presents an important avenue for further longitudinal studies with pre-literate children (Goswami, 2015a).

2.2.1.3. Automaticity. To explain both language and non-language based difficulties reported amongst dyslexics, Nicolson, Fawcett, and colleagues proposed deficits in automatizing skills are causal of dyslexia (Nicolson & Fawcett, 1990, 1999, 2011; Nicolson, Fawcett, & Dean, 2001). This theory is associated with the limited capacity view described in Chapter 1 and suggests that, like many other complex skills, reading and writing become automatized during development to allow for fluent execution and to free up cognitive resources for higher level processes (see p. 21). The automaticity deficit theory posits that deficits in automatizing skills result in atypical reading and spelling development.

Nicolson and Fawcett (1990) tested this theory using a dual-task paradigm whereby children would maintain balance – a skill assumed to highly automatized – in one condition and then maintain balance whilst completing a secondary counting or auditory pitch discrimination task in a second condition. Their hypothesis was that if automatization was unaffected and balance was automatized than dyslexics would not be impaired in the dual-task condition. They found that children with dyslexia were impaired in the dual task but not the single balancing condition. In addition to deficits in motor skills, automatization deficits have been found in non-motor activities (Nicolson & Fawcett, 2000). These findings were

taken as support for the thesis that children with dyslexia had difficulties in automatizing skills.

Nicolson, Fawcett, and Dean (2001) identified a brain basis for the automatization deficit by establishing a link between impairments in the cerebellum and automatization deficits. The cerebellum forms part of the cortico-cerebellar system and is heavily involved in the acquisition and production of skilled motor actions (see Doyon, Penhune, & Underleider, 2003) and language processing (Booth et al., 2007). Accordingly, Fawcett, Nicolson, and Dean (1996) and Fawcett et al. (1996) reported that children with dyslexia performed significantly less well than children without dyslexia on clinical tests of cerebellar function including those of postural stability (balance), muscle tone, and complex movements (e.g., pointing, toe tapping etc.). Using neuroimaging, adults with dyslexia were found to have lower activation in the right cerebellum during a finger sequencing task (Nicolson et al., 2001).

Based on their behavioural and neuroimaging case-control studies, Nicolson and Fawcett (1999; 2011) proposed a causal model to explain several impairments commonly found amongst children with dyslexia, including phonological deficits. According to this model (see Figure 2.1), cerebellar deficits led to behavioural impairments in writing, reading, and spelling following three separate paths. In the writing path, the authors suggested that cerebellar deficits led to motor difficulties, which in turn led to writing difficulties. A second path suggests that deficits in the cerebellum/cortico-cerebellar tracts lead to articulation deficits, in turn this leads to deficits in phonological processing and predominantly reading, but also spelling difficulties. A third path suggests that automatisation deficits also lead to spelling difficulties.

Overall, an automatization theory offers a holistic explanation of the heterogeneous profiles found amongst individuals with dyslexia. Furthermore, the model described by

Nicolson and Fawcett (1999) offer some theoretical explanation to writing difficulties described in dyslexic samples (e.g., Berninger et al., 2008; Connelly, Campbell, & Maclean, 2006). However, there are several limitations to the automatization deficit hypothesis in both the causal model and in this theory more generally. In several areas, the model described in Nicolson and Fawcett (1999) is either under specified or has since been falsified. For example, the authors refer to difficulties in writing, however, it is not clear to what aspect of writing they were referring. It is most likely that they were referring to handwriting, due to the pathway involving motor skills. As discussed earlier, however, handwriting is not just related to motor skills but spelling too (e.g., Abbott & Berninger, 1993), yet this link is not acknowledged. The model also implies a separate basis for the development of reading and spelling, yet a large literature now reports that reading and spelling are likely to share the same bases (e.g., Caravolas et al., 2012).
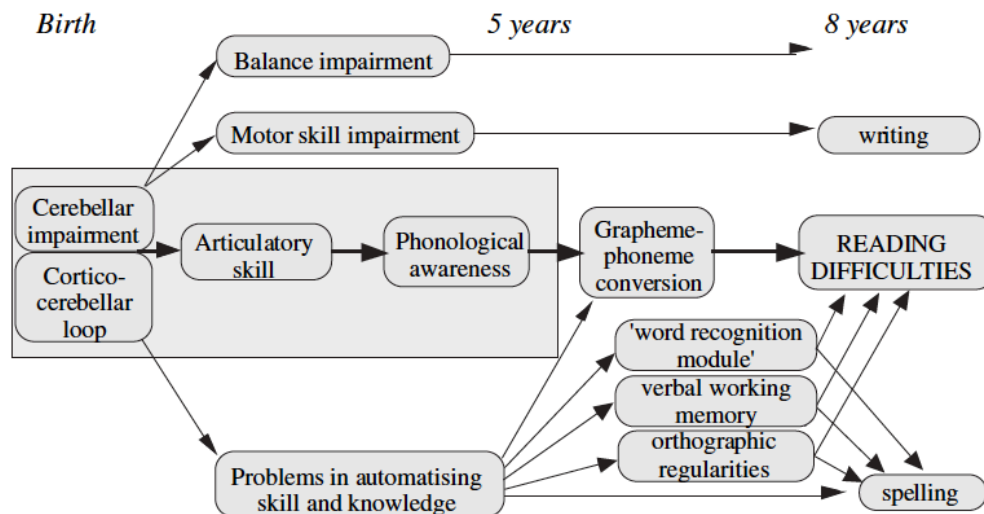
Figure 2.1. Nicolson and Fawcett's (1999; 2011) model of causal relationships between cerebellar dysfunction, automatization, and dyslexia.

Several limitations weaken the automatization theory more generally. Chiefly among the limitations is that most of the evidence presented by Nicolson, Fawcett and colleagues

appears to come from the same cohort of children (see Nicolson & Fawcett, 1999). Few other research groups using different samples have replicated findings of cerebellar/automatization deficits. Wimmer, Mayringer, and Landerl (1998) compared children with and without dyslexia using a similar dual-task balancing paradigm to that reported by Nicolson and Fawcett (1990) as well as measures of fine motor, phonological, and articulation skills. The authors found no differences on the dual-task paradigm, fine motor skills, or articulation skills but dyslexics were impaired on measures of phonological skills, suggesting children with dyslexia did not exhibit an automatization deficit but did have deficits in phonological processing. Wimmer et al. (1998) noted that children with ADHD often had balancing difficulties – which led to children with ADHD being excluded in their study – and suggested that the reason for the difference between their findings and those reported by Nicolson and Fawcett (1990) could stem from children with comorbid dyslexia and ADHD in the original sample.

In a follow-on study, Wimmer, Mayringer, and Raberger, (1999) did not exclude children based on attentional difficulties but took teacher ratings of ADHD instead. Initially, they found a balancing deficit in the dyslexia group but when they removed children with high ratings of ADHD, they found no balancing deficit amongst children with dyslexia. Furthermore, there was a strong correlation between ADHD ratings and dual-task balancing performance. Thus, the dual-task difficulties – which form the basis for the automatization hypothesis – appear to reflect ADHD more strongly than dyslexia. This conclusion was echoed in a meta-analysis of balance impairments in dyslexia which also found balance deficits were more strongly related to comorbid disorders such as developmental coordination disorder (DCD) and ADHD (Rochelle & Talcott, 2006).

The automatization deficit hypothesis explains typical as well as atypical development as it is necessary to automatize skills. However, outside the Nicholson and

Fawcett research group there is little evidence to substantiate their claims. Furthermore, there is little evidence that children with dyslexia have established automatization deficits before learning to read, or, that automatization deficits explain most of the behavioural impairments of the disorder.

      2.2.1.4. Summary. Dyslexia is a neurodevelopmental disorder that affects the acquisition of reading and spelling skills. Several theories have proposed causal deficits of dyslexia. Amongst these theories, a phonological, visual attention span, and automatization deficits have all been proposed as causal of dyslexia. Of the theories reviewed here, the phonological and visual attention span hypotheses have the theoretical capacity to explain typical and atypical development. There is also strong evidence of a longitudinal relationship between phonological processing deficits and later reading and spelling impairments. Evidence from training studies demonstrates remediation of phonological processing deficits improves reading and spelling, providing support for a causal role of phonological processing. Yet, such support for the visual attention span deficit hypothesis has not yet come to fruition. Furthermore, phonological processing deficits are present in the majority of individuals with dyslexia. Thus, phonological processing should be considered a core deficit of dyslexia. However, as is apparent, even this core deficit does not explain all cases of dyslexia. Moreover, a core deficit of phonological processing does not account for the high comorbidity between dyslexia and other neurodevelopmental disorders such as DCD.

2.2.2. Developmental coordination disorder (DCD)

      The labels dyspraxia, developmental coordination disorder, and developmental motor coordination disorder have all been applied to individuals who have impairments in the development and use of coordinated motor skill. In this thesis, the term and classification proposed by the Diagnostic Statistical Manual of Mental Disorders (DSM-V; APA, 2013) is followed. In the DSM-V, developmental coordination disorder (DCD) is broadly defined by

four criteria. A label of DCD is applied where *"acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use. Difficulties are manifested as clumsiness, as well as slowness and inaccuracy of performance of motor skills"* (Criterion A; APA, 2013, p. 74). These motor difficulties interfere with academic and daily activities (Criterion B), occur early in development (Criterion C), and are not explained by any neurological or physiological disorder (Criterion D).

The diagnostic criteria described above are non-specific in relation to the types of coordinated motor skill difficulties that would be defined as DCD. For example, an individual with a selective balance problem would be eligible to meet the DSM-V criteria as well as an individual with fine motor difficulties. The non-specificity of the diagnostic criteria makes DCD a very heterogeneous disorder; more so than dyslexia.

Despite the common prevalence and the continued impact DCD has on those with the disorder, it is one of the least researched neurodevelopmental disorders (Bishop, 2010). There is a wide range of neurophysiological, cognitive, and behavioural impairments associated with DCD (Wilson et al., 2017). This has resulted in several competing hypotheses regarding its aetiology, yet a unifying theory remains elusive (Gomez & Sirigu, 2015). In the main, theories of DCD stem from a cognitive, cognitive neuroscience, or ecological systems perspective (Wilson, Ruddock, Smits-Engelsman, Polatajko, & Blank, 2013). Here, I restrict my overview to theories from a cognitive and cognitive neuroscience perspective and briefly discuss hypotheses regarding perceptuomotor and internal modelling deficits.

2.2.2.1. Perceptuomotor processing. Visual and kinaesthetic mapping is utilised in learning motor actions, particularly prior to establishing motor programmes/sensorimotor maps (Halsband & Lange, 2006; Wolpert & Ghahramani, 2000). In the context of motor action, visuospatial processing is important for localising and object identification

(Jeannerod, 2006). For example, visual information is important for feeding back when writing along a line to ensure the written trace stays on the line. Kinesthesis describes the awareness of our body movements. It is often conflated with proprioception (an awareness of static body position) in the literature (Hill, 2005) and so for parsimony kinesthesis and proprioception are referred to as kinesthesis here. Kinaesthetic information is supplied via muscular, joint, and skin receptors and provides a reference of where limbs are in space (Jeannerod, 2006). Here, I refer to visual, kinaesthetic, and cross-modal integration of sensory information used in motor-related processes as *perceptuomotor processes*, but they have also been referred to as sensorimotor processes. Deficits in processing this information have been proposed as being causally related to DCD (Wilson & McKenzie, 1998).

    2.2.2.1.1. Visuospatial processing. Visuospatial processing entails a complex system that involves identifying and recognising objects (Jeannerod, 2006;Valyear, Culham, Sharif, Westwood, & Goodale, 2006)). Children with DCD appear to have diffuse deficits in all aspects of visuospatial processing (Sigmundsson, Hansen, & Talcott, 2003). Early work by Hulme and colleagues (Hulme, Smart, & Moran, 1982; Lord & Hulme, 1987, 1988) found children with DCD were impaired on tasks involving judging visuospatial relationships between simple stimuli (e.g., line length judgement and size discrimination of simple objects). Moreover, large correlations between visual discrimination performance and drawing errors amongst children with DCD were present indicating a relationship between impaired visuospatial processing and functional motor impairments (Lord & Hulme, 1988).

    Since the early work by Hulme and colleagues, several studies have also found children with DCD to have impaired visuospatial processing abilities (Parush, Yochman, Cohen, & Gershon, 1998; Schoemaker et al., 2001; Tsai, Wilson, & Wu, 2008; Tsai & Wu, 2008; Wilson & McKenzie, 1998). In the main, these studies utilise a case-control design and compare children with and without DCD on batteries of tests that are assumed to tap various

visuospatial processing abilities (e.g., Test of Visual Perception Skills; TVPS; Gardner, 1996). Some measures require no motor output and are referred to as visuoperceptual tasks whilst others require motor action and are often referred to as complex visuospatial tasks (Wilson & McKenzie, 1998). Separate meta-analyses have reported large impairments relative to controls on both visuoperceptual and complex visuospatial tasks (Wilson & McKenzie, 1998; Wilson et al., 2013). Thus, deficits in visuospatial processing appear to be large and pervasive in DCD.

Whilst visuospatial processing deficits are common amongst children with DCD, the locus of these deficits is not clear. There is considerable heterogeneity at the group level of children with DCD on tasks measuring visuospatial processing. For instance, in Schoemaker et al. (2001), 20% of the DCD group scored less than 1.5 *SD* on a task of matching rotated figures. Yet, 47% of the DCD sample scored less than 1.5 *SD* when matching figures that were incomplete. In addition, others have completely failed to find any differences between children with and without DCD on visuospatial tasks or to replicate earlier findings (Bonifacci, 2004; Schoemaker et al., 2001). This has led to some questioning whether the task demands moderate performance (Wilson et al., 2017).

Similarly, inconsistencies in the relationship between performance on visuospatial processing tasks and motor performance has been noted (Prunty, Barnett, Wilmut, & Plumb, 2016; Schoemaker et al., 2001). Henderson, Barnett, and Henderson, (1994) failed to find a significant correlation between visuospatial processing and drawing errors when replicating Lord and Hulme (1988). However, others have reported significant associations between visuospatial processing and motor skill measures. Tsai and Wu (2008) reported correlations between measures of manual dexterity and measures of visuospatial memory and perception and balancing. It is important to note that these findings were correlational, and the presence or absence of correlations does not infer the presence or absence of causality.

Possible explanations for the inconsistent findings include the differing diagnostic categories employed by studies of DCD and the reliance on case-control designs. As mentioned earlier, the broad criteria of identifying DCD produce very heterogeneous samples of children with DCD, which could explain the inconsistent findings found in these studies. The use of case-control designs is also pervasive throughout the literature on DCD (see Wilson et al., 2013). Whilst these designs are useful for establishing deficits amongst individuals with DCD, the design does not afford the ability to confirm or reject a causal relationship between visual processing deficits and DCD. Although a visuospatial processing deficit has the capacity to explain typical and atypical development it remains unclear whether the presence of such deficits influence the development of later motor skills and whether it can explain most of the behavioural impairments of the DCD.

2.2.2.1.2. Kinaesthetic processing. An alternative perceptual explanation of DCD is a kinaesthetic processing deficit. As noted earlier, kinaesthetic information is important for monitoring and updating during skilled motor action (Jeannerod, 2006). A kinaesthetic deficit was initially promoted by Bairstow, Lazlow and colleagues (Bairstow & Laszlo, 1981; Laszlo & Bairstow, 1983; Laszlo, Bairstow, Bartrip, & Rolfe, 1988) and later by Coleman, Piek, and Livesey (2001) and Li, Su, Fu, and Pickett (2015).

To identify kinaesthetic deficits, Lazlow and Bairstow (1985) developed the Kinaesthetic Sensitivity Test (KST). The test was composed of two components, the *kinaesthetic acuity* (discriminating passive arm movements) and *kinaesthetic perception and memory* (memory and mapping of complex movement patterns with visual information) subtests. Using this test, Bairstow and Lazlow (1981) reported eight out of fourteen children with DCD had kinaesthetic processing deficits (Bairstow & Lazlow, 1981). Later, Lazlow et al. (1988) used the KST both as a pre- and post-test measure as well as part of the intervention for improving motor difficulties amongst children with DCD. Unsurprisingly,

children who received the KST training performed significantly better at post-test, which the authors cited as evidence for a causal relationship between kinaesthetic deficits and DCD. A more likely explanation is that training the KST improved subsequent performance on the KST. This position has been supported by subsequent intervention studies which have failed to find improved motor coordination after kinaesthetic training (e.g., Polatajko et al., 1995).

In addition, the reliability and validity of the KST has been questioned (Gomez & Sirigu, 2015; Visser & Geuze, 2000). The KST has been found to have poor discriminative validity (Smyth & Mason, 1997) and it is likely that the task taps heavily on other processes including integration of perceptual and motor skills (Coleman et al., 2001). Thus, the concerns of validity of the KST and the lack of an association between kinaesthetic ability and motor skills raises questions regarding a causal link between kinaesthetic processing and functional motor impairments found in DCD.

2.2.2.1.3. Cross-modal integration deficits. A third aspect of perceptuomotor deficits associated with DCD is the cross-modal integration of perceptual information. Cross-modal integration has been referred to as integration between several different perceptual modalities, but most commonly it is discussed in the context of visual and kinaesthetic integration. Accordingly, case-control studies have also reported that children with DCD perform less well than typically developing controls on tasks requiring cross-modal integration (Hulme et al., 1982; Mon-Williams, Wann, & Pascal, 1999). However, these deficits were only small to moderate in size (Wilson & McKenzie, 1998; Wilson et al., 2013) and no significant associations have been found between cross-modal integration and motor skills.

To recap, perceptuomotor processes play a vital role in motor production, particularly in the acquisition of skilled motor action. Deficits of visuospatial, kinaesthetic, and, cross-modal processing has been reported amongst children with DCD. Researchers have proposed

separate causal links between visual processing deficits and DCD and kinaesthetic deficits and DCD. Whilst both explanations have the potential to explain motor development, there is a lack of empirical evidence of causality between these deficits and DCD. In the case of kinaesthetic deficits, the training study lauded as demonstrating a causal link between kinaesthetic ability and DCD was methodologically flawed and concerns have been raised in the measurement of kinaesthetic ability (Siringu & Gomez, 2015). There is more consistent evidence for visuospatial processing deficits amongst children with DCD (Wilson & McKenzie, 1998; Wilson et al., 2013). However, robust longitudinal and intervention studies of visuospatial processing and motor skills are necessary for establishing a causal relationship.

    2.2.2.2. Internal modelling deficit (IMD). An alternative hypothesis from a cognitive neuroscience perspective suggests that DCD results from deficits in utilising forward internal models (Wilson et al., 2004). Forward internal models are an important aspect of the motor system. When motor commands are generated by the motor cortex they are sent to motor effectors. In addition, an *efference copy* or *corollary discharge* of the motor command is also generated and sent to the parietal and cerebellar cortices. The efference copy acts as a forward internal model of the predicted outcome of the motor action and provides several functions. The forward internal model is compared with sensory feedback to detect and correct a mismatch in the predicted and actual state. In addition, forward internal models provide stability to the motor system prior to slow perceptual (visual and kinaesthetic) feedback. In this case, the internal model provides feedback of the predicted outcome prior to slower perceptual feedback and is described as predictive control. Internal forward models also accommodate learning by predicting the outcomes of actions without executing them, thus allowing rehearsal of the actions (Wolpert, 1997; Wolpert & Ghahramani, 2000).

According to the IMD hypothesis, deficits in utilising internal models during motor learning and action leads to the motor coordination deficits of DCD (Wilson et al., 2004). Converging evidence in support of this hypothesis comes from paradigms measuring motor imagery. Motor imagery is a representation of a motor action without motor output and is assumed to reflect the efference copy of motor commands (Crammond, 1997; Gomez & Sirigu, 2015). Common paradigms used to assess motor imagery include mental rotation tasks. Case control designs have found that children with DCD typically perform significantly less well than children without DCD on these tasks (Wilson et al., 2004; Wilson et al., 2013; Wilson, Thomas, & Maruff, 2002).

Mental rotation tasks involving the rotation of limbs are considered to be valid and reliable measures of motor imagery. Wilson et al. (2004) used a mental rotation of hands to examine motor imagery amongst 16 children with and 18 children without DCD. Hands were presented to children at 45° intervals between 0 – 180°. Children were required to decide whether the hand was a right or left hand. Typically developing children performed as expected on this task where their reaction time increased linearly with increasing angle of the hand (Shepard & Metzler, 1971). However, for children with DCD, reaction time increased at a slower rate than typically developing children. The authors argued the smaller trade-off between reaction times and angle of rotation reflected impairments in using motor imagery appropriately. These conclusions are in accordance with other work finding atypical performance by children with DCD on mental rotation tasks (Williams et al., 2011; Williams, Thomas, Maruff, & Wilson, 2008; Wilson et al., 2004).

Children with DCD have also been found to perform less well than typically developing children on tasks requiring predictive control including pointing (Lewis, Vance, Maruff, Wilson, & Cairney, 2008), reach-to-grasp (Smyth & Mason, 1997), and load-lifting (Jover, Schmitz, Centelles, & Chabrol, 2010). Indeed, a meta-analysis of these tasks found

that children with DCD had large impairments relative to controls on tasks tapping forward internal modelling/predictive control (Wilson et al., 2013). The strong converging evidence has led some to suggest deficits in forward internal modelling are a core impairment of DCD (Wilson et al., 2013).

Whilst it is clear that children with DCD have deficits in forward internal modelling, a causal relationship between these deficits and DCD remains unverified. Much of the evidence presented in favour of the IMD is based on low powered case-control designs (Adams, Lust, Wilson, & Steenbergen, 2014; Wilson et al., 2013). For example, in Adams et al.'s (2014) systematic review, the average sample size of children with DCD was 20.4 children. Aside from making generalisations difficult, small sample sizes make it hard to test associations between the IMD and functional motor skills.

To my knowledge, there are currently no longitudinal studies examining the relationship between internal modelling ability and later motor skills (Adams et al., 2014). As discussed earlier, such studies are an important piece of evidence in establishing causality. In addition to the lack of longitudinal designs, only one intervention study has examined the effects of training internal modelling on motor skills amongst children with DCD. The training study compared computer motor imagery training with perceptuomotor training and no training in children with DCD. Motor imagery and perceptuomotor training led to significant gains in scores on a standardised motor assessment battery (Wilson et al., 2002). However, a third of children identified with DCD in this study had motor skills in the low normal range making it hard to ascertain whether the intervention would have the same effect amongst children with clinical motor impairments. Further studies training motor imagery in children who meet the full diagnostic criteria for DCD are necessary to establish a causal link between IMD and DCD.

2.2.2.3. Summary. DCD is a neurodevelopmental disorder that affects the acquisition of coordinated motor skills. Like dyslexia, several theories have proposed that specific deficits are causal of DCD. This short review focused specifically on deficits in perceptuomotor and internal modelling processes. These theories have the capacity to explain typical and atypical development of motor skills. Furthermore, when compared with typically developing children, children with DCD have large deficits in visuospatial processing and internal modelling (Wilson & McKenzie, 1998; Wilson et al., 2013). Whilst these deficits appear to be large, studies demonstrating deficits in visuospatial and/or internal modelling processes as causally related to DCD remain elusive. Also, neither the perceptuomotor nor IMD hypotheses account for the high frequency of comorbidity between DCD and other neurodevelopmental disorders, particularly dyslexia.

2.2.3. Multifactorial View of Neurodevelopmental Disorders

The brief review of cognitive deficits associated with dyslexia and DCD highlighted that no single deficit could fully account for the heterogeneous profiles of dyslexia and DCD. In addition, none of the theories reviewed in the previous section could explain the high frequency of comorbidity between dyslexia and DCD (see Study 2, pp. 142 - 149 for extended discussion on the relation between the two disorders). These two issues are not unique to dyslexia and DCD but to all neurodevelopmental disorders and have led to reconceptualising developmental disorders from single factor to a multifactorial view (Bishop, 2006; Pennington, 2006).

Pennington (2006) proposed the multiple deficit model (MDM) to explain the multifactorial nature of complex behaviourally defined neurodevelopmental disorders like dyslexia and DCD. The model was originally developed to account for the aetiologies and comorbidities between dyslexia and speech sound disorder (SSD) and dyslexia and ADHD. This means that the model can account for homotypic (comorbidity between disorders within

the same domain, e.g., SSD and dyslexia) as well as heterotypic (comorbidity between different domains, e.g., ADHD and dyslexia; Angold, Costello, & Erkanli, 1999) comorbidity. The model explains the mechanisms of disorders at four levels, the aetiological, neural, cognitive, and symptom. Each of the four levels will now be discussed in turn.

At the highest, aetiological level, multiple genetic and environmental factors interact and correlate with one another. Genetic and environmental factors act in a probabilistic manner to influence neural and cognitive development. They can either be risk factors by increasing the chance of deficits at the neural and cognitive levels or protective by decreasing the risk of deficits downstream (van Bergen, van der Leij, & de Jong, 2014).

Genetic and environmental factors influence the development of neural systems. Note that a singular genetic or environmental factor can influence the development of multiple neural systems (defined as pleiotropy). There is also further interaction at the neural level where the development of one system influences another. This means that atypical development in one system may also affect the development of other systems. In turn, the development of neural systems affects the development of cognitive processes. The development of cognitive processes is also highly interactive. Therefore, atypical neural development leads to atypical development in cognitive processes, which also has a knock-on effect on the development of other processes. Multiple cognitive deficits lead to symptoms of disorder(s) at the symptom level.

The MDM makes five assumptions about neurodevelopmental disorders including that the (a) aetiology of disorders is multifactorial resulting from multiple risk and protective genetic and environmental factors, (b) risk and protective factors influence the development of cognitive processes in development resulting in symptoms at the symptom level, (c) disorders do not result from a single aetiological factor, (d) comorbidity is likely due to shared cognitive and aetiological factors, (e) the likelihood of a disorder occurring is

continuous and quantitative making the threshold for impairment arbitrary (Pennington, 2006).

Assumptions of the MDM raise two important points worthy of further thought. The first is that multiple aetiological factors are necessary for a disorder. This means that theories which propose a single deficit is responsible for a disorder – like those reviewed separately for dyslexia and DCD – are unlikely. Rather, multiple deficits are necessary to induce the behavioural impairments which accumulate to meet diagnostic thresholds for a disorder (Hulme & Snowling, 2009). This means that cognitive deficits should be considered as *cognitive risk factors* where the presence of a risk factor increases the likelihood of meeting a diagnostic threshold for a disorder, but the presence of a risk factor alone does not mean the individual would be diagnosed with the disorder. In this view, phonological and visuospatial deficits could be considered as cognitive risk factors for dyslexia and DCD, respectively.

The MDM also suggests that some aetiological and cognitive risk factors are shared between disorders making comorbidity likely between disorders from different domains. This assumes that comorbidity between developmental disorders reflects the same aetiology as non-comorbid expressions (singular disorders) of the disorders. Crucially this assumption can be directly tested by comparing children with singular and comorbid disorders. In addition, the model differentiates between two types of risk factors, those that are specific to a disorder and those that are shared between disorders.

Under the multifactorial approach, considerable progress has been made in examining profiles of performance and identifying independent and shared cognitive risk factors of comorbidity between, dyslexia, ADHD, language impairment, and dyscalculia (de Jong, Oosterlaan, & Sergeant, 2006; Gooch, Hulme, Nash, & Snowling, 2014; Gooch, Snowling, & Hulme, 2011; Moll, Göbel, Gooch, Landerl, & Snowling, 2016). However, to date, no study has tested whether the MDM holds true in explaining the comorbidity between dyslexia and

DCD nor has any study examined shared and independent cognitive risk factors between these disorders. As such, the hypothesised MDM for dyslexia and DCD is presented in Figure 2.2. Understanding the relationship between dyslexia and DCD is vital for understanding shared impairments in transcription skills amongst these disorders.

Level of Analysis

Aetiological risk and protective factors

G_1 E_1 G_2 E_2 G_3

Neural systems

N_1 N_2 N_3

Cognitive/motor processes

$C_{Phonological}$ $C\text{-}M_{Percpetuomotor}$ C_3

Behavioral disorders

$D_{dyslexia}$ D_{DCD} D_3

Spelling   Handwriting
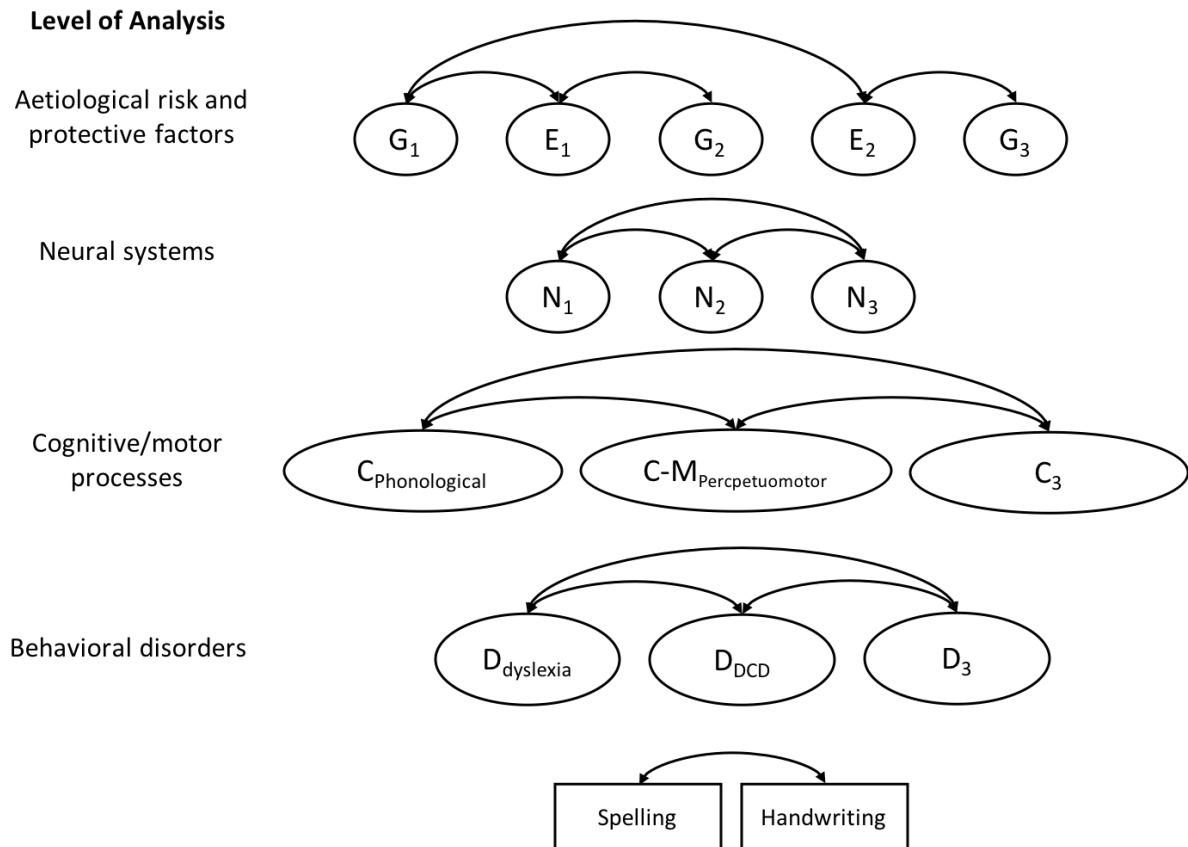
Figure 2.2. Adapted version of Pennington's (2006) multiple deficit model (MDM) to explain dyslexia and DCD. Multiple genetic (G) and environmental (E) risk and protective factors interact to influence the development of neural systems (N). The development of neural systems interacts and affect the development of cognitive (C)/cognitive-motor processes (C-M). Cognitive development overlaps and interact with one another. Multiple deficits lead to the development of complex behaviourally defined disorders (D) such as dyslexia and DCD. Double headed arrows represent interactions.

2.3. Transcription Impairments in Dyslexia and DCD

The review thus far has focused on two rather distinct literatures, the typical development of writing – particularly in relation to the development of transcription skills – and the literatures describing dyslexia and DCD, two common neurodevelopmental disorders which are frequently comorbid with one another. It remains to bring together these two distinct literatures by evaluating research that reports a high degree of overlap in spelling and handwriting impairments in dyslexia and DCD.

2.3.1. Spelling Impairments in Dyslexia and DCD

As noted earlier, proficient spelling requires knowledge of multiple linguistic constructs. Spelling impairments have been reported separately amongst children with dyslexia and children with DCD. As such, studies describing these impairments in each disorder are discussed separately.

2.3.1.1. Dyslexia. In English, spelling difficulties often accompany reading difficulties amongst children with dyslexia, and spelling difficulties have been included in some definitions of dyslexia (c.f. Rose, 2009). As discussed earlier, phoneme awareness is critical for the growth of spelling skills (e.g., Caravolas et al., 2001; Caravolas et al., 2012). Deficits in phonological processing, including phonological awareness, have been causally associated with dyslexia (e.g., Hulme et al., 2012). It is therefore logical that impairments in spelling skills amongst children with dyslexia are related to phonological processing deficits. Indeed, children with dyslexia make more spelling errors that are phonologically implausible suggesting children with dyslexia have difficulties in applying phonological knowledge to the spellings (Caravolas & Volín, 2001).

2.3.1.2. DCD. Spelling impairments have also been reported amongst children with DCD. In a large cohort study of 6959 children, children with DCD (scoring within the 5th centile of an adapted version of the Motor Assessment Battery for Children; M-ABC;

Henderson & Sugden, 1992) were found to be significantly poorer spellers than typically developing children. Similar reports have also found children with DCD have significantly poorer spelling ability than children without DCD (Alloway, 2007; Archibald & Alloway, 2008; Dewey, Kaplan, Crawford, & Wilson, 2002). Alloway (2007) reported that 46% of children with DCD achieved literacy scores < 1 *SD* below their age average on a composite measure of reading and spelling skills.

Unlike dyslexia, there is no strong theoretical or empirical link between deficits in spelling related processes and DCD. There are however three possible explanations for poorer spelling skills in DCD. The first is that dyslexia and DCD are different expressions of the same disorder. According to Nicolson and Fawcett (2007), dyslexia, DCD, developmental language disorder (DLD), and ADHD are different manifestations of a *procedural learning deficit* resulting from disruption in the cortico-striatal and cortico-cerebellar pathways. However, this explanation seems unlikely as children with dyslexia and children with DCD have intact procedural learning (Biotteau, Chaix, & Albaret, 2015).

An alternative explanation is that impairments in spelling amongst children with DCD are a secondary consequence of motor skill impairments. In a similar vein to the Mathew's effect in reading, children with DCD who have handwriting difficulties resulting from their primary motor deficits are less likely to practice writing and specifically, spelling. This means that they might have less opportunity to build phonological to orthographic mappings and have lower lexical exposure, resulting in poorer word spelling ability. Currently, there is no direct evidence to support a secondary association between DCD and spelling.

A third, explanation is that poorer spelling found amongst children with DCD is a result of comorbid cases in the group. It follows that dyslexia and DCD are frequently comorbid with one another (Cruddace & Riddell, 2006; Kaplan et al., 1998). Despite the high risk of comorbidity none of the studies that found spelling impairments amongst children

with DCD, reported whether they had excluded children with dyslexia. This opens the possibility that at least some of the sample had comorbid dyslexia resulting in decreasing the group average spelling scores. Evidence to support this assertion comes from Alloway (2007) who found a proportion of her sample to have significant spelling difficulties and a large variability of spelling ability found in DCD groups (Archibald & Alloway, 2008; Dewey et al., 2002).

2.3.2. Handwriting Impairments in Dyslexia and DCD

Developing fluent and legible handwriting is important for the development of higher-level writing skills (e.g., McCutcheon, 2000). Handwriting fluency and – to a lesser extent – handwriting legibility difficulties have been described in both dyslexia and DCD. In comparison to spelling impairments, fewer studies have examined handwriting impairments in dyslexia and DCD. Here, I briefly review the studies describing handwriting fluency and legibility difficulties amongst children with dyslexia and DCD. The reader is referred to Study 3 (pp. 205 – 214) for a more comprehensive overview of the literature pertaining to handwriting difficulties in dyslexia and DCD as this literature is directly relevant to the research questions addressed there.

2.3.2.1. Fluency. The advent of pen tracking technology has facilitated our understanding of dysfluent handwriting in children with neurodevelopmental disorders. Recent work has revealed that dysfluent handwriting is present in both dyslexia and DCD across a variety of writing tasks (Berninger et al., 2008; Kandel et al., 2017; Prunty et al., 2013; Prunty, Barnett, Wilmut, & Plumb, 2014; Rosenblum & Livneh-Zirinski, 2008; Rosenblum, Margieh, & Engel-Yeger, 2013; Sumner et al., 2013, 2014).

Studies by Sumner and colleagues (Sumner et al., 2013, 2014; Sumner, Connelly, & Barnett, 2016) reported children with dyslexia were no different from age-matched controls in the speed with which they wrote but they paused more frequently, reducing their overall

handwriting fluency. Children with dyslexia made significantly more pausing errors within words with spelling errors (Sumner et al., 2016). Furthermore, pausing durations as well as spelling ability – but not fine motor skills – explained 76% of the variance in overall handwriting fluency amongst dyslexics (Sumner et al., 2014). In a separate study children's verbal fluency and executive functions (inhibition/task switching) – but not fine motor skills – explained some variance (19.3%) in handwriting fluency (Berninger et al., 2008). It is therefore clear that children with dyslexia have poorer handwriting fluency and these impairments are associated with spelling and – to a lesser extent – with executive functions rather than fine motor ability.

Children with DCD are more dysfluent writers across a range of writing tasks. Like findings amongst children with dyslexia, children with DCD have been found not to differ from typically developing children on handwriting speed, but do make more frequent and longer pauses, thus reducing children's overall fluency (Prunty et al., 2013, 2014). Further analyses of atypical pausing behaviours revealed children with DCD made longer pauses within words that were illegible rather than words that were spelt incorrectly. Pauses within words were predicted by the child's fine motor, but not literacy, skills suggesting the locus of handwriting difficulties was related to motor deficits (Prunty et al., 2014).

The weight of the evidence suggests both children with dyslexia and children DCD have similar types of handwriting fluency impairments – namely atypical pausing behaviours – which are related to different mechanisms. Atypical pausing behaviours amongst children with dyslexia are associated with spelling ability whereas atypical pausing behaviours in DCD are associated with motor ability (Prunty et al., 2013; Sumner et al., 2013). Both authors interpret these outcomes within a limited capacity view and attribute their findings to a lack of automaticity in spelling for dyslexia and motor skills for DCD.

However, there are several issues which should be resolved before coming to this conclusion. Theoretically, it is unclear how atypical pausing is linked to automaticity in these disorders and greater specification of the nature of handwriting fluency is needed to address this. At a methodological level, in the studies reported by Prunty et al. (2013, 2014) children with DCD had significantly lower spelling ability than typically developing children. Looking at the group level, spelling ability amongst children with DCD were in the average range but within the DCD group 30% of children scored below 1 *SD* on a standardised measure of spelling ability, indicating children with comorbid dyslexia and DCD may have been present in the group. The potential presence of comorbid dyslexia and DCD make it difficult to interpret whether handwriting fluency impairments are related to dyslexia or to DCD and to conclusively determine whether handwriting difficulties between these two disorders are related to dissociable impairments.

2.3.2.2. Legibility. Comparatively fewer studies have examined the profiles of handwriting legibility in dyslexia and DCD. Of those studies which have, most are concerned with legibility amongst children with DCD and less so in dyslexia.

Studies of handwriting legibility amongst children with dyslexia are predominantly limited to qualitative or descriptive analyses of the author's general impression of legibility (Berninger et al., 2008; Cooke, 2002; Martlew, 1992). For example, Martlew (1992) described how letter formation in children with dyslexia was like younger children's formation. To my knowledge, no study has systematically examined handwriting legibility amongst children with dyslexia nor examined the associations between handwriting legibility and literacy and motor ability.

Profiles of handwriting legibility have been examined a little more systematically amongst children with DCD. These studies have employed the use of global rating scales such as the Handwriting Legibility Scale (Barnett, Prunty, & Rosenblum, 2018). Using these

scales, children with DCD appear to have diffuse legibility difficulties including problems in letter formation and spatial aspects of formation (Barnett et al., 2018; Rosenblum & Livneh-Zirinski, 2008). The use of these global measures restricts a fuller understanding of handwriting impairments found in children with DCD because global measures do not allow researchers to identify if any specific aspects of handwriting legibility are impaired amongst children with DCD. Furthermore, global scores tend not to correlate well with other measures (c.f. Graham et al. 1998) making it difficult to examine associations between handwriting legibility and related skills (literacy, motor, and executive functions).

There is a dearth of research examining handwriting legibility profiles amongst children with dyslexia and children with DCD. To date, no study has attempted to examine and compare the full profiles of handwriting legibility difficulties or examine the associations between legibility ability and handwriting related skills. Knowledge of handwriting legibility profiles in dyslexia and DCD has important implications for understanding handwriting impairments in these disorders. Particularly in elucidating the relationships between deficits related to dyslexia and DCD and the nature of handwriting difficulties in these disorders.

To summarise, although recent work focusing on handwriting fluency has suggested handwriting impairments are different in dyslexia and DCD, the presence of comorbid cases in the DCD samples and the lack of direct comparisons between dyslexia and DCD groups make firm conclusions difficult. Further knowledge of the types of handwriting legibility difficulties between dyslexia and DCD would also address questions regarding the nature of handwriting difficulties in these disorders.

2.4. Conclusion

This review has brought together literatures mapping out theories of dyslexia and DCD and describing spelling and handwriting impairments in these disorders. The review has revealed many outstanding questions regarding handwriting impairments in both dyslexia and

DCD. Despite being separable disorders, there is considerable overlap in spelling and handwriting impairments reported amongst dyslexia and DCD. Whilst spelling impairments are expected in children with dyslexia, impairments have also been reported amongst children with DCD. In addition, dysfluent handwriting marked by frequent pausing has been reported in both dyslexia and DCD. An issue with current studies of spelling and handwriting in dyslexia and DCD is the potential uncontrolled comorbidity between the two disorders. As spelling and handwriting skills are important foundation skills for writing (e.g., Berninger & Amtmann, 2003; Berninger & Winn, 2006), understanding spelling and particularly handwriting impairments amongst children with dyslexia, DCD, and comorbid dyslexia and DCD is vital for improving our knowledge of how these impairments affect writing development.

2.5. The Current Study

The primary goal of this thesis was to understand the nature of writing difficulties in children with dyslexia, DCD, and comorbid dyslexia and DCD. Having established that transcription skills play a fundamental role in learning to write, but are often impaired in both dyslexia and DCD, it is important to understand the nature of these impairments in relation to specific disorders. A complicating factor in this pursuit is that the relationship between the two disorders themselves is not clear and raises its own set of questions. In this thesis, I take a programmatic approach to address two related aims. The first aim was to understand the relationship between dyslexia and DCD. The second aim was to understand the nature of transcription difficulties in dyslexia and DCD. In the first half of this thesis I addressed the first aim by examining the relationship between dyslexia and DCD (Studies 1 and 2). In the second half of this thesis (Studies 3 and 4) I addressed aim two by examining the nature of transcription – particularly handwriting difficulties – between dyslexia, DCD, and comorbid dyslexia and DCD.

2.5.1. Overview of Studies

    2.5.1.1. Study one. Prior to examining transcription skills in dyslexia and DCD, it was first necessary to understand the relationship between dyslexia and DCD. A primary issue in understanding the relationship between dyslexia and DCD was to establish whether the disorders were frequently comorbid. The multiple deficit model (MDM) proposes that comorbidity between dyslexia and DCD should be expected (Pennington, 2006). Although evidence from clinical samples and small-community samples do suggest comorbidity between the two disorders is higher than expected by chance, no study has examined whether comorbidity of dyslexia and DCD is greater than expected by chance in a large community-based sample. We addressed this primary aim in Study 1. Finding that the frequency of comorbid dyslexia and DCD is greater than statistical chance would provide tentative support for the MDM and suggest that the comorbidity of dyslexia and DCD should be further investigated.

    2.5.1.2. Study two. A second issue in understanding the relationship between dyslexia and DCD was to understand areas of potential overlap between the disorders. Assuming a multifactorial view of these disorders (e.g., Pennington, 2006), we examined candidate independent and shared (phonological, perceptuomotor, and executive function) cognitive risk factors/markers of dyslexia and DCD. In addition to cognitive deficits, we investigated whether there was overlap at the symptom level between dyslexia and DCD. That is, whether children with dyslexia and DCD had overlapping impairments in literacy and motor skills. This was particularly relevant for testing whether spelling impairments amongst children with DCD reported by some (e.g., Alloway, 2007) were the result of comorbid cases or not. A final aim of this second study was to test another assumption made by the MDM regarding comorbidity. Accordingly, the MDM assumes that comorbidity is the result of shared aetiological and cognitive risk factors between disorders. According to this

view, the profiles of impairments amongst children with comorbid dyslexia and DCD should be a combination of impairments of similar severity to children with dyslexia-only and DCD-only singular disorders (Pennington, 2006). We tested this by directly comparing children with and without dyslexia-only, DCD-only, and comorbid dyslexia and DCD. Because the skill markers in this study (phonology, perceptuomotor, executive, and literacy) are also related to handwriting production, establishing the presence or absence of these deficits in dyslexia and/or DCD would suggest potential areas of association to pursue further in understanding the nature of handwriting difficulties in these disorders.

2.5.1.3. Study three. Having established the nature of the relationship between dyslexia and DCD, we focused on the nature of handwriting fluency and legibility difficulties in dyslexia and DCD. Specifically, children with dyslexia, DCD, and comorbid dyslexia and DCD's handwriting fluency and legibility profiles were compared in detail. It was hoped that directly comparing group profiles on multiple measures would enable a deeper understanding of the nature of handwriting difficulties in these groups and identify dissociations in difficulties between the groups. Finding dissociations between groups in the performance on handwriting measures and different patterns of correlations between handwriting performance and handwriting related skills was indicative of separate mechanisms of handwriting impairments between dyslexia and DCD.

2.5.1.4. Study four. The largest handwriting legibility impairments found in both dyslexia and DCD were those in forming letters. Learning to form letters is a complex process requiring children to learn letter's motor programmes and phonological forms. In this final study, we examined whether handwriting impairments in dyslexia and DCD were related, in part, to learning motor programmes of the letters. To examine motor programme learning and control for potential confounds such as knowledge of the letter's phonological

form we used a training study in which children with dyslexia, DCD, and comorbid dyslexia and DCD learned novel orthographic characters.

Chapter 3

General Methods

This thesis addressed different but inter-related questions all concerned with understanding the relationship between dyslexia and DCD and transcription impairments. The separate studies in this thesis used a variety of paradigms to address specific research questions which are described with the specific studies. However, the studies in this thesis were unified by the overall design and the same sample of children who took part. As such, the overall design is reviewed here.

3.1. Overall Design

A major challenge in researching neurodevelopmental disorders is identifying and recruiting adequate numbers of children with the disorder of interest. This challenge is amplified when examining multiple single and comorbid disorders, as was the case in this thesis. To mitigate some of the challenges with recruitment, we used a screening and assessment design. Many children from local schools ($N = 733$) were initially screened for markers of literacy (dyslexia), motor (DCD), and literacy and motor (comorbid dyslexia and DCD) difficulties or no markers of both difficulties. Children who met the criteria for having markers of literacy and/or motor difficulties as well as children without any markers were then invited to take part in the rest of the project. During the rest of the project, children completed several sessions of assessments and tasks that formed the last three studies of this thesis.

In addition to children screened and assessed from local schools, children from local specialist educational (Miles Dyslexia Centre and Conwy Learning support service) and clinical (NHS occupational and physiotherapy clinics) services were referred to the project. These children were all attending mainstream school in the community and were tested at their school or at the university.

The overall design used here mitigated the issues in recruiting samples of children with dyslexia, DCD, and comorbid dyslexia and DCD for every study undertaken. Arguably,

this design is more powerful as it affords the chance for comparisons across the studies. For example, it enabled skills investigated in Study 2 in the context of the disorders dyslexia and DCD to be also examined in relation to handwriting impairments in Studies 3 and 4. The size of the studies both in terms of the sample sizes and of the number of measures administered meant that the studies were spread over 18 months. The duration of participation meant that this thesis was longitudinal in nature, however, examining development over time was not of direct interest here. The design of this thesis can be broken down into three phases and is graphically outlined in Figure 3.1.

3.1.1. Phase One

In Phase 1 (Summer 2015) 733 children in Years 3, 4, and 5 took part in Study 1.

3.1.2. Phase Two

In Phase 2 (Winter 2015 – Summer 2016), 140 children who were now in Years 4, 5, and 6 were identified as having markers of literacy and/or motor difficulties or were typically developing children were invited to continue participating. At this point, approximately 17 children referred from clinics (Miles Dyslexia Centre, Local Education Authority Teaching Service, and NHS Occupational Therapy Services) but were in full time mainstream education in North Wales joined the study. By the end of Phase 2, children completed most of the measures for Study 2 and all the tasks for Study 3 (see Appendix A for full breakdown of the time point specific tasks were administered). At Phase 2, the children in Year 6 also completed Study 4 because they were due to graduate from their primary education that summer.

3.1.3. Phase Three

Finally, in Phase 3, children in Years 4 and 5 at Phase Two were now in Years 5 and 6. They completed the rest of the measures administered as part of Study 2 and the

experimental tasks for Study 4 (see Appendix A for full breakdown of the time point specific

tasks were administered).
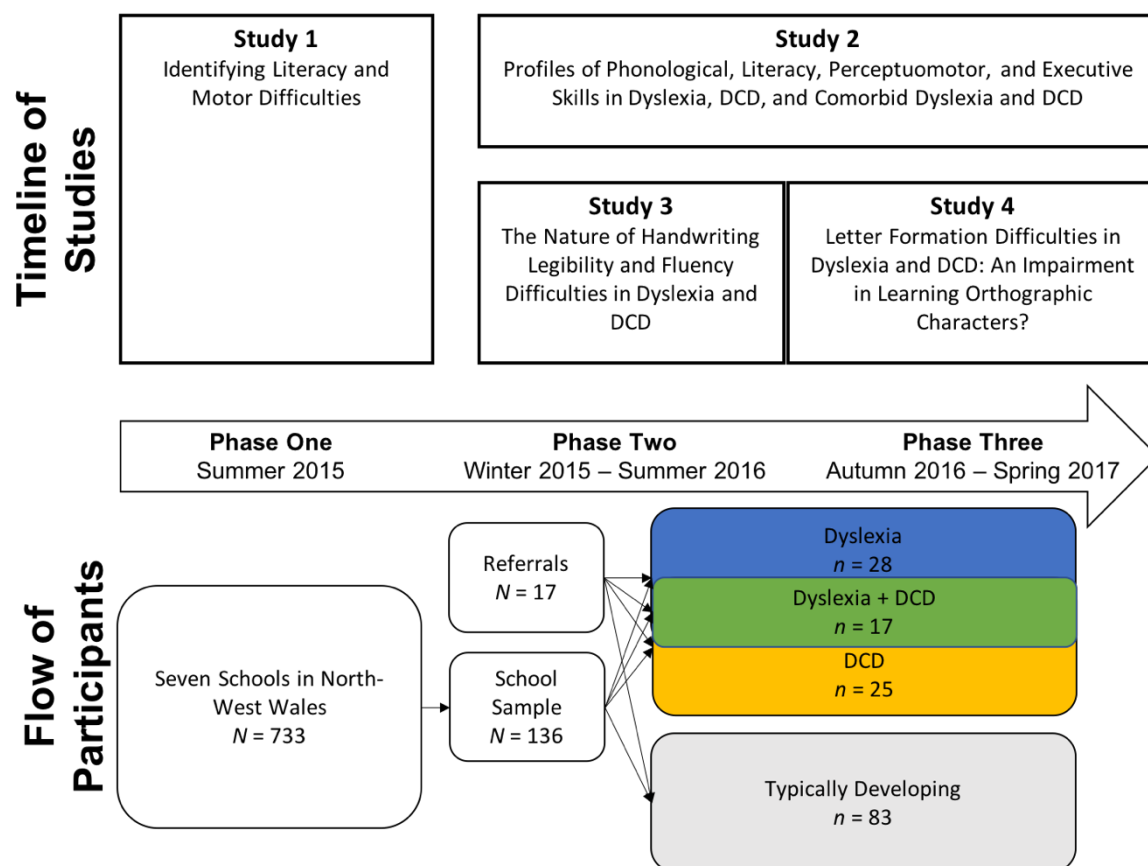
Figure 3.1. Overview of the studies contained within thesis including a timeline of when

testing was completed for each study and a breakdown of the flow of participants.

3.2. Ethical Considerations

The studies contained within this thesis were reviewed and approved by the School of

Psychology's Research Ethics Committee at Bangor University (reference number: 2015-

15287) and an NHS Research Ethics Committee (reference number: 16/WA/0141). Fully

informed written consent was gained from head teachers for the entire project. For Phase 1,

parents were informed of the project and what it would involve for their child. As is common

in psycho-educational studies such as this, parental consent was assumed via opt-out at this

stage, with head teachers also assuming loco-parentis. From Phase 2, and in the case of all

children referred to the project via specialist services, informed written consent was gained

from parents. Verbal consent was gained from every child during individual testing in Phases 2 and 3.

During Phases 2 and 3, children were rewarded after completing each task with positive affirmation and stickers. All tasks were short in length, not exceeding 20 minutes in length, with breaks between them. Where standardised tests were used, published administration procedures were followed. If a child complained of being tired or did not wish continue testing was stopped immediately. After each session, children were verbally debriefed. Following data collection and analyses, schools received research reports of aggregated data of pupil performance on several measures most relevant to educators. Parents were also provided with individualised data pertaining to their child on request.

Chapter 4

Study 1: Identifying Literacy and Motor Difficulties in a Non-Selected School

Population: Prevalence and Comorbidity

4.1. Introduction

According to multifactorial theories of neurodevelopmental disorders (Pennington, 2006; Thapar & Rutter, 2015) the high frequency of comorbidity between disorders such as dyslexia and DCD is the result of shared aetiological risk factors. Evidence to support this view partly comes from large scale epidemiological studies of disorders which demonstrate that the frequency of comorbidity is greater than would be expected by chance, based on the rates of singular disorders (Caron & Rutter, 1991). However, no investigation has examined the prevalence of dyslexia and DCD from large community-based samples. Doing so is important as both dyslexia and DCD share overlap of some impairments such as handwriting difficulties (Berninger et al., 2008; Prunty et al., 2013). Therefore, an investigation is necessary to establish whether the frequency of comorbidity between dyslexia and DCD is greater than expected by chance. In turn, such a finding has implications for future investigations of transcription difficulties amongst children with dyslexia, DCD, and co-occurring dyslexia and DCD.

4.1.1. Multifactorial Models of Neurodevelopmental Disorders

As discussed in Chapter 2, the current view is that the aetiology of neurodevelopmental disorders – a heterogeneous group of disorders which include ADHD, developmental language disorder, speech sound disorder, dyslexia, dyscalculia, DCD, and autism spectrum disorder – is multifactorial in nature (Thapar & Rutter, 2015). To account for the multifactorial nature of these disorders Pennington (2006) proposed a complex behavioural model over four levels: aetiological, neural, cognitive, and symptom. At the aetiological level, complex interactions between environmental and genetic risk and protective factors influence the development of multiple neural systems, either at the same time, or during later development. Neural systems affect the development and action of

multiple cognitive processes which interact with one another. The impairments at the cognitive level lead to symptoms of the disorder(s) at the symptom level.

An advantage of this model over alternative single-deficit models is that it explains the highly comorbid nature of neurodevelopmental disorders (Pennington, 2006) where comorbid disorders are the result of shared aetiological and cognitive risk factors. Evidence for this multifactorial account of comorbid disorders comes primarily from investigations of comorbidity in dyslexia and ADHD. Investigations have found shared genetic (e.g., Willcutt et al., 2002) and cognitive (e.g., Gooch, Snowling, & Hulme, 2011; Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005) deficits amongst children with dyslexia, ADHD, and comorbid dyslexia and ADHD in both clinic and community-based samples (Germanò, Gagliano, & Curatolo, 2010).

A crucial aspect of establishing a shared aetiology between disorders is to determine whether the frequency of comorbid disorders is greater than that predicted from the base rates of singular disorders. If the frequency of children with comorbid disorders is greater than the frequency predicted from the combined frequency of singular disorders it can be concluded that the two disorders are related. However, analysing the frequency of comorbid disorders in a clinic-based sample leads to artificially inflated prevalence estimates (see Caron & Rutter, 1991). To gain a true estimate of comorbidity it is important to estimate the prevalence of both singular disorders and of comorbid cases from a large representative sample (Caron & Rutter, 1991).

No study has investigated the prevalence of dyslexia, DCD, and comorbid dyslexia and DCD among a large community-based sample. However, such community-based samples have been used to estimate the prevalence of dyslexia-only and DCD-only separately. Studies estimating the prevalence of singular dyslexia and DCD are reviewed briefly before work examining comorbid dyslexia and DCD is discussed.

4.1.2. Identification and Prevalence of Dyslexia

Dyslexia, defined as a learning difficulty affecting the skills used in accurate and fluent word reading and spelling (Rose, 2009), affects between 3 – 10% of the English speaking population (Snowling, 2013). The variation in prevalence estimates is due to differences in how dyslexia is defined and identified (Snowling & Hulme, 2015), including the use of different cut-offs. For example, in a population-based study of 2586 Austrian children, Landerl and Moll (2010) applied a 1 *SD* cut-off and found 14.8% and 16.4% of the sample to have reading and spelling difficulties, respectively. However, when they applied a more conservative cut-off of 1.5 *SD* the prevalence rates dropped by half to 7% and 8.8% for reading and spelling difficulties, respectively.

The most recent estimate of reading difficulties, or dyslexia, amongst a large sample of children from the United Kingdom (UK) comes from the standardisation sample of the York Assessment of Reading Comprehension (YARC; Snowling et al., 2009). During the standardization, 662 children in primary schools completed the four subtests of the YARC. Using a 1.5 *SD* cut-off on the YARC's Single-Word Reading Test, Snowling and Hulme (2015) reported 10.5% of the sample as having significant reading difficulties, suggesting the current UK prevalence of dyslexia is likely to be at the upper end of general estimates.

Studies which estimate the prevalence of dyslexia such as Snowling et al. (2009) and others (Rutter et al., 2004; Shaywitz, Shaywitz, Fletcher, & Escobar, 1990) have been vital for mapping the prevalence of dyslexia in English speaking children. These studies have exclusively focused on identifying children with reading difficulties rather than those with reading and spelling difficulties. Only identifying children with reading difficulties is in part due to historical focus on reading impairments only. However, both reading and spelling skills share the same cognitive predictors of development including phoneme awareness (Caravolas et al., 2012). Deficits in phoneme awareness are considered a hallmark feature of

dyslexia which also cause spelling difficulties (Caravolas & Volín, 2001). Moreover, spelling difficulties are often harder to remediate than reading difficulties (Romani, Olson, & Betta, 2005). Therefore, identifying both reading and spelling impairments could lead to more sensitive prevalence estimates of dyslexia. Prevalence studies examining dyslexia and comorbid neurodevelopmental disorders have identified frequent comorbidity between dyslexia and several other disorders including ADHD (Willcutt & Pennington, 2000), SLI (Catts, Adlof, Hogan, & Weismer, 2005), dyscalculia (Dirks, Spyer, van Lieshout, & de Sonneville, 2008), and DCD (e.g., Cruddace & Riddell, 2006).

4.1.3. Identification and Prevalence of DCD

The most frequently cited prevalence estimates of DCD in the literature range between 5 – 6% (Blank, Smits-Engelsman, Polatajko, & Wilson, 2012). Prevalence estimates of DCD are affected by several related factors which include the criteria used for identifying DCD, the aetiology of the disorder, and associated measurement issues. DCD is broadly defined in the current revision of the DSM-V (APA, 2013) by four criteria. These criteria were described in Chapter 2 (p. 51) but are revised briefly here. A label of DCD is given when the action of co-ordinated motor skill is below the expected standard considering the individual's age and opportunity for learning the skill (Criterion A), motor difficulties interfere with academic and daily activities (Criterion B), occur early in development (Criterion C), and are not explained by any neurological or physiological disorder (Criterion D).

The broad classification criteria for DCD in the DSM-V reflect the largely unknown aetiology of DCD (Zwicker, Missiuna, Harris, & Boyd, 2012) and the broad range of motor skills which are tested in norm referenced tests. Common norm referenced tests which include the Movement Assessment Battery for Children 2 (M-ABC 2; Henderson, Sugden, & Barnett, 2007) and the Bruiniks-Oseretsky Test of Motor Proficiency (BOTMP-2; Bruininks

& Bruininks, 2005) often measure motor ability during simple tasks which tap different aspects of motor skill. However, differences exist between the tests on which aspects of motor skills are tested. For instance, the BOTMP-2 explicitly measures strength and agility whereas the M-ABC 2 measures coordinated motor accuracy during tasks of manual dexterity (fine motor), aiming and catching (gross motor), and balance. As these tests are often used to operationalise Criterion A of the DSM, it is likely that the prevalence of DCD will differ according to the test used.

Current norm-referenced tests of motor skills also rely on observational scoring by the administrator. Observational scoring introduces subjectivity and makes it harder to objectively measure motor ability and reduces the reliability of the test, which in turn increases the variability of prevalence estimates. The variability in the motor skills measured between tests and the reliance on observational scoring are two possible explanations of the low agreement found between norm-referenced tests of motor skills (Crawford, Wilson, & Dewey, 2001). As such, no gold standard of motor assessment exists (Mcintyre et al., 2017) and identification of DCD often varies between studies (Smits-Engelsman, Schoemaker, Delabastita, Hoskens, & Geuze, 2015).

Due to the observational nature of many norm referenced motor tests, they must often be administered to children on a 1:1 basis which makes assessing large groups of children – as is important for prevalence studies – resource and time intensive. Alternatives to behavioural methods include parent/teacher questionnaires (e.g., the M-ABC Checklist Teacher Questionnaire) which are useful for assisting with a clinical diagnosis, but not for screening due to their poor sensitivity (Barnett, Hill, Kirby, & Sugden, 2015; Blank et al., 2012).

Studies examining prevalence of DCD have attempted to mitigate the aforementioned methodological and logistical issues by using behavioural measures with two-step screening

and assessment approaches (e.g., Wright & Sugden, 1996). Despite the difficulties described earlier, one comprehensive epidemiological study, conducted by Lingam, Hunt, Golding, Jongmans, and Emond (2009) as part of the Avon Longitudinal Study of Parents and Children (ALSPAC), has investigated the prevalence of DCD using the diagnostic criteria described in the DSM-IV (APA, 1996). The ALSPAC is a population-based study investigating the health and development of 6,959 children who were due to be born between 1st April 1991 and 31st December 1992 and whose mothers resided in Avon, UK.

The DSM-IV largely corresponds with the current criteria of DSM-V. However, it placed a greater emphasis on excluding children with low IQ under Criterion D. Nevertheless, the DSM-IV criteria for DCD were operationalised by Lingham et al. (2009) as scoring below the 15th centile on three representative tests of the MABC (Criterion A). Criterion B was operationalised as scoring below the 10th centile on a parent questionnaire of activities of daily living, and below the national average on a curriculum based handwriting test. Although, the authors qualify neither whether handwriting legibility or fluency was assessed/nor the details of the scoring system. DCD was not identified in cases where neurological/medical impairments influenced motor development (Criterion C) or in children with an IQ < 70 on the WISC-III (Criterion D). Using these criteria, Lingam et al. (2009) found 4.9% of the sample had DCD which corresponds to the frequently reported prevalence estimates between 5 – 6% (Blank et al., 2012).

Children who were identified as having DCD in the ALSPAC cohort were subsequently found to be at an increased risk for handwriting, reading, attentional, and non-verbal difficulties (Schoemaker, Lingam, Jongmans, van Heuvelen, & Emond, 2013). The finding of an increased risk of comorbid difficulties amongst children with DCD supports other clinic- and community-based studies which have found high rates of comorbid

difficulties between DCD and ADHD (Rasmussen & Gillberg, 1996), SLI (Hill, 2001), ASD (Lingham et al., 2009), and dyslexia (Cruddace & Riddell, 2006).

Accurate estimates of comorbid dyslexia and DCD are difficult to establish due to a lack of representative, community-based, prevalence studies using behavioural measures which identify both singular and comorbid dyslexia and DCD. The lack of such studies likely reflects the methodological difficulties associated with identifying DCD. However, studies have attempted to mitigate these methodological issues by using clinic or small community-based samples.

4.1.4. Current Prevalence Studies of Comorbid Dyslexia and DCD

Much of the work investigating the prevalence and profiles of comorbid dyslexia and DCD has used clinic-based samples (e.g., Dewey, Kaplan, Crawford, & Wilson, 2002; Kaplan, Dewey, Crawford, & Wilson, 2001). An early clinic-based investigation undertaken by Kaplan et al. (1998) assessed motor, reading, and attention skills in 224 children referred to the authors for having learning or attention difficulties, along with 155 controls who had no reported difficulties. Owing to the issues in testing motor skills, the authors administered multiple motor assessments. Accordingly, children were tested on the BOTMP (Bruinicks, 1978), Movement Assessment Battery for Children (M-ABC; Henderson & Sugden, 1992), and DCD Questionnaire (DCDQ; Wilson, Kaplan, Crawford, Campbell, & Dewey, 2000). Children were identified as having DCD if they scored below the recommended thresholds reported in each of the test's manuals for identifying impairments. These criteria identified 50% of the sample as having DCD.

Broad criteria were used for assessing reading ability, whereby reading accuracy and comprehension were assessed using a battery of standardised tests including the Woodcock-Johnson Psychoeducational Battery-Revised (WJ-R; Woodcock & Johnson, 1989), Wide Range Achievement Test-Revised (WRAT-R; Jastak & Wilkinson, 1984), and the Auditory

Analysis Test (Rosner & Simon, 1971). Reading disabilities, or dyslexia, were identified if participants met any one of the following three criteria (a) they scored below 16th centile on the Basic Reading aspect of the WJ-R, (b) below the 15th centile on Reading Comprehension tests from the WJ-R, (c) below the 30th centile on the WJ-R Word Attack test/ below the 16th centile on the WRAT-R or WJ-R Spelling Tests, and below 16 on the AAT. Using these broad criteria, 43.8% of the sample were identified as having dyslexia.

Of those who met the criteria for either DCD or dyslexia, 33% fulfilled the criteria for both disorders, suggesting a third of the sample had comorbid motor and reading difficulties. This high rate of comorbidity is somewhat surprising as no child was referred to the study for having motor difficulties. However, the high rates reported in this study are likely to be inflated due to recruitment of a clinic sample which means prevalence rates are likely to be inflated (Caron & Rutter, 1991). The estimates are also likely to be inflated due to the use of broad criteria for identifying disorders, particularly in reading disabilities.

In addition to studies of clinic-based samples, investigations have attempted to assess prevalence of comorbid dyslexia and DCD in small community samples using parent/teacher questionnaires (e.g., Martin, Piek, Baynam, Levy, & Hay, 2010) or hybrid combinations of questionnaires followed up with behavioural assessments (Cruddace & Riddell, 2006). Cruddace and Riddell (2006) screened 129 children aged between 9 and 10 years old by collecting teacher reports of each child's reading and spelling, motor, and attention skills. Based on teacher identification, 68 children completed a behavioural battery and were categorised as having a reading difficulty if their Word Reading score was significantly below their General Conceptual Ability score on the British Ability Scales II (BAS II; Elliott, 1996) and/or if they scored below the 10th centile on the Nonword Graded Reading test (Snowling, Stothard, & MacLean, 1996). A child was identified as having motor difficulty if they scored below the 5th centile on the M-ABC.

To establish prevalence estimates in their sample, the authors compared the number of children categorised as having dyslexia and/or DCD with the total number of children originally screened using teacher reports (129). A total of 15 children (21%) met the criteria for reading difficulty, 16 children (23%) for motor difficulty, and 9 children (13%) for reading and motor difficulty. The frequency of reading and motor difficulties were below that reported by Kaplan et al. (1998) but more than double that would be expected based on the rates of the singular disorders. Like Kaplan et al. (1998), Cruddace and Riddell's (2006) data suggest an increased risk of comorbid literacy and motor difficulties.

Cruddace and Riddell (2006) report the prevalence estimates of comorbid reading and motor difficulties based on their sample probabilities and not population probabilities. However, the high incidence of singular reading (11.6%) and motor (12.4%) difficulties in their sample were inflated in comparison to the commonly reported population prevalence rates. Reasons for these very high rates of singular reading and motor difficulties may include the relatively small sample size for an epidemiological study or the use of teacher report questionnaires which are ill advised for identifying reading and motor difficulties (Barnett et al., 2015; Blank et al., 2012; Shaywitz et al., 1990). The disproportionate frequency of singular literacy and motor difficulties found in the sample raises questions about the accuracy of the base rates. Accurate base rates are necessary for determining whether the frequency of co-occurring literacy and motor difficulty is greater than chance alone (Caron & Rutter, 1991). To conclude that there is an increased risk of comorbidity between dyslexia and DCD it is necessary to examine the frequency of comorbid difficulties in a sample where the rates of singular dyslexia and DCD are similar to population prevalence estimates.

No study has been able to estimate the prevalence of comorbid dyslexia and DCD using representative base rates of singular dyslexia and DCD. However, Schoemaker et al. (2013) established an increased risk of reading difficulties in a representative sample of

children with DCD. In a follow on study with the ALSPAC cohort, Schoemaker et al. (2013) examined whether children who were identified as having DCD by Lingham et al. (2009) were at greater risk of additional difficulties including reading, spelling, and handwriting. In addition to the motor skills assessment described earlier, children were identified as having additional difficulties if they performed below the 10th centile on assessments of reading (Wechsler Objective Reading Dimensions; WORD; Rust, Golombok, & Trickey, 1993) and spelling.

Children with DCD were found to be at a significantly greater risk of having reading, but not spelling, difficulties than controls. As such, Schoemaker et al. (2013) demonstrated an increased risk of comorbid reading difficulties amongst children with DCD identified using behavioural measures in a large community sample. Although Shoemaker et al. (2013) did not examine the number of children with reading but not motor difficulties in the same sample, these findings, along with those from Kaplan et al. (1998) and from Cruddace and Riddell (2006) suggest an increased risk of comorbidity between dyslexia and DCD.

4.1.5. The Current Study

The importance of establishing the prevalence of singular and comorbid dyslexia and DCD in a representative community sample is clear. In this study, a screening approach was used to achieve this end. A screening approach affords the ability to assess a large sample of children in order to get indicative prevalence estimates of children at risk of singular and comorbid dyslexia and DCD. Due to difficulties in assessing motor skills (as discussed earlier) and given that concurrent screening of literacy and motor skills has not been undertaken before, one aim here was to establish whether behavioural tasks administered as part of a screening battery were suitable for assessing literacy and motor skills.

To facilitate screening large groups of children for DCD, we opted to use tests of perceptuomotor (e.g., visual motor integration) and handwriting skills. Whilst

perceptuomotor skills are utilised in all types of skilled motor action (Halsband & Lange, 2006) the tasks used in this study are arguably those most related to fine-motor skills and less so to other aspects of motor skills such as gross motor skills or balance. Unfortunately, methods for testing gross motor and balance skills with large numbers of children in the classroom remain elusive. It therefore remains an open question as to whether the perceptuomotor and handwriting measures used in the current study were able to detect children with broader motor coordination difficulties in addition to those with difficulties in fine motor skills.

It is also important to note that screening approaches should not be used for diagnoses. Therefore, children whom we deem to be at risk of having dyslexia will be identified as having literacy difficulties. Children whom we deem to be at risk of having DCD will be identified as having motor difficulties. Those who we deem to be at risk of comorbid dyslexia and DCD will be identified as having comorbid literacy and motor difficulties.

We expect the prevalence of singular literacy and motor difficulties to match the rates of dyslexia and DCD described in the literature. The prevalence estimates of singular and comorbid literacy and motor difficulties will be used to establish whether the frequency of comorbid literacy and motor difficulties is greater than that expected by chance alone. Based on existing research on comorbidities among developmental disorders (e.g., Gooch et al., 2011; Moll, Göbel, Gooch, Landerl, & Snowling, 2016), we anticipated to find the frequency of comorbid literacy and motor difficulties to be greater than expected based on the rates of singular difficulties; such an outcome may suggest a shared aetiology between literacy and motor difficulties.

4.2. Methods

4.2.1. Participants

A total of 733 children were screened in Years 3 (n = 239, M age = 8.2 years, SD = 0.53, 51% female), 4 (n = 257, M age = 9.1 years, SD = 0.54, 49% female), and 5 (n = 237, M age = 10.2 years, SD = 0.53, 48% female) from seven primary schools across North-West Wales (see Table 4.1 for breakdown of sample size by year group and school).

All but one school were classified by the Welsh government as either 'predominantly English medium with significant use of Welsh' or 'predominantly English medium' (Welsh Government, 2007). At foundation phase, children in these schools were taught in both languages with a greater emphasis on English, or, exclusively in English. At key stage two, children were taught predominantly through the medium of English. Only one school in the current study was categorised as a Welsh-medium school (Welsh Government, 2007). Children in this school were taught through the medium of Welsh at foundation phase and continued to be instructed in Welsh for at least 70% of the curriculum at key stage two. However, the Welsh Government (2007) expects children in Welsh medium schools to reach a standard of English equivalent to children in English medium schools by the end of Year 6. In addition, children who attended the Welsh medium school, were more likely to have Welsh as their first language (see Table 4.1).

Table 4.1.

Sample Sizes by Year Group and Language Profiles of Children in Each Participating School

	Year Group (*n*)				Home Language – L1(%)		
School	3	4	5	Total	English	Welsh	Other
Sch1	60	58	50	168	95	0	5
Sch2	30	29	30	89	88	0	12
Sch3	39	46	41	126	94	0	6
Sch4	30	28	28	86	93	0	7
Sch5	19	17	20	56	96	0	4
Sch6	30	36	35	101	94	4	2
Sch7[a]	31	43	33	107	67	33	0

Note. [a]Primary language of instruction was Welsh.

4.2.2. Design and Procedure

The primary aim of the current study was to examine the prevalence of literacy, motor, and comorbid literacy and motor difficulties in a community-based sample. As such, whole classes of children completed all the measures described below in specially prepared booklets, and the screening was completed during normal class activities. For ease of administration and to reduce fatigue effects, measures were delivered over two 60-minute sessions (see Table 4.2 for session and administration order of tasks).

All sessions were administered by the candidate and two or three trained research assistants; this was done to maintain good oversight of children's needs during task completion. Both sessions were completed within one week of each other and in many cases on the same day. When administered on the same day, the sessions were separated with a break of at least an hour. Children were told at the beginning of the first testing session that the sessions were not like normal tests and more like games, but they should try their best and work individually.

Table 4.2

Administration Order of Tests, the Skills they Measured, and their Corresponding

Hypothesised Skill Domains

Test	Session	Order	Measuring	Skill Domain
Matrices	1	a	Non-verbal ability/ visuospatial	Perceptuomotor
Welsh Reading		b	Welsh reading	
Welsh Spelling		c	Welsh spelling	
Coding		d	Graphomotor speed	Perceptuomotor
Word Spelling	2	a	English word spelling	Literacy
Cloze Reading		b	English reading	Literacy
Sentence Dictation		c	English sentence spelling Handwriting legibility	Literacy Handwriting
Visual Motor Integration		d	Visuospatial	Perceptuomotor

Note. No hypothesised skill domain is given for the Welsh tasks because these were control measures and so were not used to identify markers of difficulty.

4.2.3. Measures

As no dedicated test battery exists for the purpose of concurrently screening literacy and motor skills in classrooms, several established tests were selected to measure a number of literacy and motor related skills, which are described below.

4.2.3.1. Literacy. Reading and spelling skills were assessed in English and in Welsh. English Reading, Word Spelling, and Sentence Spelling tests were used to measure English Literacy skills and identify children who had literacy difficulties who were therefore at risk of dyslexia. Measures of Welsh reading and spelling (see below) were taken as an additional indirect indicator of the level of Welsh instruction delivered by each school.

4.2.3.1.1. English reading. Reading proficiency (accuracy and comprehension) was assessed using a timed, cloze sentence completion task (Caravolas & Volín, 2001).

Participants were instructed to read short passages with either one or two missing words, and to select the missing word(s) needed to complete the sentence as quickly as they could. Each missing word was selected from five options including the target item and four phonologically and semantically plausible distractor items. Passages varied in length between 7 to 45 words and increased in difficulty by length, vocabulary, and general knowledge. The test comprised thirty items: in the first 14 passages, one word was missing whilst in the latter passages, two were missing. Where two words were missing, participants selected the first missing word from five choices presented in a list labelled 'A' and the second missing word from a choice of five words from a list labelled 'B'. The score was the number of correctly identified target words within eight minutes with a maximum score of 46.

4.2.3.1.2. English word spelling. The Wide Range Achievement Test IV (WRAT-IV; Wilkinson, & Robertson, 2006) Spelling subtest was used as a measure of single word spelling in which participants spelt 13 alphabet letters followed by 36 words graded in difficulty. The cut-off of 36 words was used based on the expectation that most of the oldest children were unlikely to achieve a standard score exceeding 145. The administrator read the words aloud three times. Once in isolation, a second time within a carrier sentence and then in isolation once more. Participants were given approximately 30 seconds to write each word. Scoring followed published guidelines in which a correct spelling was awarded one point and scoring was discontinued after 10 consecutive errors. The maximum possible score was 49.

4.2.3.1.3. English sentence spelling. Sentence writing was measured using the Sentence Dictation task from Caravolas, Volín, and Hulme (2005) which now forms the spelling subtest of the Spelling and Handwriting Legibility Test (SaHLT; Caravolas & Downing, in prep.). Ten graded sentences were read aloud to the class by the administrator and children were instructed to write down the sentence exactly as they heard it. Each sentence was read once prior to the participants beginning to write and then was repeated by

the administrator at short intervals until all participants had finished writing. Sentence length ranged from four to eight words with a total of 62 words presented across the 10 sentences. Words varied in phonological, morphological, lexical, and graphotactic difficulty. Each correctly spelt word was awarded one point regardless of capitalization or punctuation errors. Spelling accuracy was the sum of correctly spelt words. The maximum score was 62.

4.2.3.1.4. Welsh reading. Welsh word reading proficiency was assessed using a Welsh adaptation of the Picture Word Matching task (Caravolas, Lervåg, Defior, Seidlová Málková, & Hulme, 2013). Children matched a picture with one of four printed word choices. Word choices consisted of one target item, a phonographically similar, semantically similar, and an unrelated distractor. Children were instructed to read each set of words carefully before selecting the item which best matched the picture. The test was administered for three minutes, and the score was the number of correctly identified target words, out of a possible 63, within 3 minutes.

4.2.3.1.5. Welsh spelling. Children's Welsh spelling knowledge was assessed using a Welsh word spelling test similar to the English word spelling test (Caravolas, 2010). Children wrote 15 letters to dictation before writing 36 words, which increased in difficulty. Letters and words were pre-recorded by a native Welsh speaker for a standardized delivery across administrators. Each word was played to the class once in isolation, a second time within a carrier sentence, and a final time in isolation. The spelling score was the total number of correct letters and words with a maximum possible score of 51.

*4.2.3.2. **Motor Skills.*** As discussed earlier, the screening of motor skills presents many challenges. This is in part due to the broad criteria used to define DCD, the differences between tests in the aspects of motor skills they measure, and the use of observational assessments in these tasks that require children to be assessed individually. To identify children at risk of motor difficulties in this study we measured their perceptuomotor skills

which are often impaired amongst children with motor difficulties (see Wilson, Ruddock, Smits-Engelsman, Polatajko, & Blank, 2013). We combined perceptuomotor tests with measures of handwriting skills as handwriting abilities have been used previously to fulfil criterion B of the DSM (APA, 2013) diagnosis of DCD (Lingam et al., 2009). Moreover, children with handwriting difficulties are of particular interest in this thesis. Perceptuomotor skills were measured using tests tapping visuospatial skills. Graphomotor skills were measured using a speeded coding test, and handwriting skills were measured using a bespoke handwriting legibility test.

    4.2.3.2.1. Visual Motor Integration. The Beery-Buktenica Developmental Test of Visual Motor Integration (Beery VMI; Beery & Beery, 2010) provided a standardized measure of visuospatial processing which can be administered individually or in groups. Children copied a series of 24 forms of increasing complexity displayed in sets of three in their booklets. Participants were asked to copy the shapes, exactly as they saw them, into the box below without using any additional aids (rulers, protractors, etc.). Only one attempt was allowed per form and no time limit was imposed. Scoring followed published guidelines in which each correctly copied item was awarded one point and scoring ended after three consecutive attempts scoring zero.

    4.2.3.2.2. Matrices. The Wide Range Intelligence Test (WRIT) Matrices subtest (Glutting, Adams, & Sheslow, 2000) was adapted for group administration and used as a measure of visuospatial skill and non-verbal ability. Children identified and selected the missing 'piece' from several possible distractors to complete a picture array.

    There were 42 test items of increasing difficulty used in this adaptation; this number was based on the expectation that many of the oldest children were unlikely to surpass this number of items without meeting the criterion for discontinuation (the achievement of which would correspond to a standard score exceeding 145). The test items were composed of four

to six incomplete pictures/patterns and these, along with three to five numbered distractor items were projected to the class onto a screen using a projector. Modified arrays were reproduced in the participants' booklets with numbered boxes replacing the choices (see Figure 4.1). Participants identified the correct missing item by circling its corresponding numbered box. In accordance with instructions in the manual, the first 35 arrays were displayed to the class for 30 seconds each, whilst the latter arrays were displayed for 45 seconds each. Scoring followed published guidelines in which participants were awarded either one or two points for identifying the correct missing element. Although children completed all 42 items, scoring was discontinued if four non-consecutive errors were made in five items, in accordance with the manual.

As this was a modified version of Matrices we sought to assess its convergent validity by correlating performance on this task with performance on another measure of non-verbal ability, Wechsler Intelligence Scale for Children-IV (WISC-IV) Block Design (Wechsler, 2004). The Block Design was delivered individually following the published administration procedure to a subset of children ($n = 40$) from the screening sample at Phase 3 (see General Methods). The correlation between the adapted Matrices and the Block Design tasks was moderate ($r = .40$, $p < .001$) indicating some convergent validity.
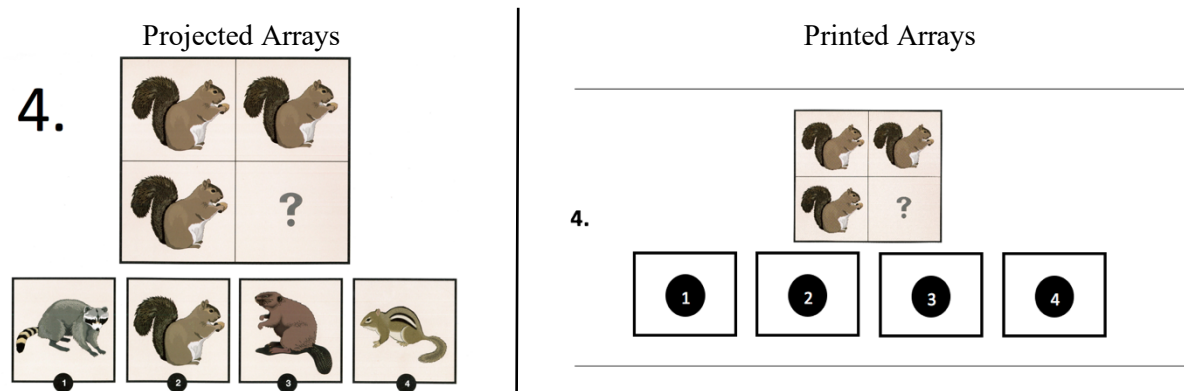
Figure 4.1. Example item arrays from the modified Matrices task. Item arrays were projected to the class with three to five distractor items beneath (projected arrays). Children made their responses by circling one of the four response options in the modified (printed) arrays reproduced in their booklets.

4.2.3.2.3. Coding. The WISC-IV Coding subtest (Wechsler, 2004) was used to index graphomotor speed. Using a numbered key of symbols printed at the top of their booklets, participants reproduced the symbols, at speed for 2 minutes, in boxes labelled with the corresponding number. The items (number-symbol pairs) were presented in a fixed pseudorandom order. Following published guidelines, responses were scored as correct if they were identifiable as the relevant symbol. The number of symbols correctly copied – out of 119 – in 2 minutes gave an index of graphomotor speed.

4.2.3.2.4. Handwriting Legibility. Responses to the Sentence Dictation task (Caravolas et al., 2005) were scored using an early version of the handwriting legibility subtest from the SaHLT, of which, the development and final version are discussed in detail elsewhere (Caravolas & Downing, in prep.). In brief, the component handwriting dimensions were theoretically motivated, guided by existing tests and national benchmarks identified from national curricular (Learning Wales, 2014; Rosenblum, Weiss, & Parush, 2003; Van Galen, 1991). The method used to score handwriting in this study is briefly described below.

Five separate scoring dimensions were applied across the ten sentences by trained assistants blind to the spelling ability of the participants. The dimensions were based on key indicators of handwriting legibility based on the theoretical and empirical literature; they include (a) *Letter Formation*, how well each letter is formed and how recognizable it is out of the context of the word; (b) *Letter Spacing*, the distance between letters within a word, specifically whether the letters are crowded or are too far apart; (c) *Word Spacing*, the distance between the words, whether the words are crowded or too far apart; (d) *Line Alignment*, how well the writing sits on the line, whether it crosses the line or 'floats' above it; (e) *Script Switching*, how consistently participants write in a cursive or script fashion. Each criterion was scored on a 7-point Likert scale ranging from 1 (fail) to 7 (excellent) with a maximum score of 35.

4.2.4. Data Analysis

Initial two-way ANOVAs were used to assess for any school clustering effects due to varying Welsh language between schools and to examine the developmental sensitivity of the tests used the battery. Relationships between literacy and motor measures were examined using correlations before factor analysis techniques were used to assess the underlying factor structure of the battery (see Table 4.2) and to identify the tests which had the greatest factor loadings from their respective constructs. The results of the correlation and factor analyses were used to select the measures for identifying children with literacy and motor difficulties. The frequency of children with literacy, motor, and comorbid literacy and motor difficulties was then examined to establish whether the frequency of comorbid literacy and motor difficulties was greater than expected by chance.

4.3. Results

The aim of the current study was to identify children with literacy, motor, and comorbid literacy and motor difficulties from a community-based sample in order to estimate the prevalence of singular and comorbid difficulties. A class screening approach was used to assess literacy and motor skills in seven primary schools in North West Wales. Initial analyses examined potential school bi-literacy clustering effects and developmental (age) sensitivity of the battery. These initial procedures informed later analyses which examined the factor structure of the battery. The factor analyses of the battery assessed the validity of the screening approach in assessing literacy and motor skills and was used to identify the most suitable tests for identifying children with literacy and motor difficulties. These tests were then used to identify children with markers of literacy, motor, and comorbid literacy and motor skills. Finally, the prevalence of literacy, motor, and comorbid literacy and motor difficulties of the sample were examined.

4.3.1. Bi-Literacy School Effects

The battery was administered in a bilingual Welsh-English region where English language use differs considerably between children within schools, by their home language use, as well between schools due to the varying amounts of instruction given through the medium of Welsh. One way to control for language effects was to recruit schools from an area where English is the predominant language. Nevertheless, confounding school-level effects arising from differences in the amount of Welsh versus English instruction across schools were possible. Potential bi-literacy effects were investigated using two-way ANOVAs with the factors of year groups and schools on performance of the reading and spelling tests administered in Welsh and English, respectively (see Table 4.3 for means and standard deviations). The ANOVAs were weighted to account for the unbalanced sample sizes in year group and school. Where significant effects of year group and school were

found on Welsh and English literacy test outcomes, follow up post-hoc comparisons using Bonferroni corrections were used to elucidate the locus of differences. Bonferroni corrected simple effects analyses were also used to assess differences between schools at each year group (see Table 4.3).

4.3.1.1. Welsh reading. Analysis of accuracy on Welsh Picture Word Matching test revealed a significant main effect of year group, $F(2, 679) = 13.79$, $p < .001$, $\eta_\rho^2 = .04$, children in Year 5 were more accurate than those in Year 4 ($p = .016$) and Year 3 ($p < .001$) and those in Year 4 were also more accurate than children in Year 3 ($p = .014$). There was also a significant main effect of school, $F(6, 679) = 17.21$, $p < .001$, $\eta_\rho^2 = .13$, pupils in the Welsh medium school, sch7, were more accurate than all other schools ($p < .006$). Furthermore, children in sch6 were more accurate than those in sch2, sch3, and sch5 ($p < .006$). Pupils in Sch1 were also more accurate than those in sch2 and sch3 ($p < .004$). There was also a significant interaction, $F(12, 679) = 2.33$, $p = .006$, $\eta_\rho^2 = .04$. Simple effects analyses revealed pupils in sch3 were significantly less accurate than children in Sch1 in Year 3 ($p = .002$), but not in older year groups. Children in sch1 and sch5 were less accurate than those in sch7 ($p < .001$) in Year 4 only. Furthermore, children in Year 4 of sch1 and Sch6 were more accurate than children in sch2. Children in sch3 were less accurate than those in sch7 in Years 4 and 5 ($p < .001$) but not Year 3. Children in Year 5 of sch7 were also more accurate than children in sch2 ($p = .029$) and sch4 ($p = .022$). Thus, there was a consistent effect of age on Welsh reading ability. Moreover, children educated through the medium of Welsh were consistently better readers than children educated through the medium of English. However, there was no consistent effects between English medium schools nor did simple effects analysis reveal any consistent patterns driving the interaction.

4.3.1.2. Welsh spelling. Welsh spelling accuracy varied according to year group, $F(2, 681) = 21.12$, $p < .001$, $\eta_\rho^2 = .06$, where pupils in Year 3 were less accurate than children in

Year 4 and Five ($p < .001$). Like Welsh reading, spelling varied according to school, $F(6, 681) = 217.72$, $p < .001$, $\eta_\rho^2 = .66$ where, as expected, pupils in sch7 were more accurate than all other schools ($p < .001$) and those in sch6 were also more accurate than sch1 – sch5 ($p < .001$). Those in sch4 were also more accurate than children in sch2, sch3, and sch5 ($p < .003$). Like Welsh reading, there was a consistent improvement of Welsh spelling with age and children educated through the medium of Welsh were better spellers of Welsh than children educated through the medium of English. Within English medium schools, children in sch6 were reliably better spellers than children in all other English medium schools. These trends likely reflect the degree to which schools deliver Welsh literacy tuition.

4.3.1.3. English reading. There were significant differences across year groups and schools in reading accuracy. The main effect of year group, $F(2, 691) = 46.08$, $p < .001$, $\eta_\rho^2 = .12$, was driven by greater accuracy in Year 5 than in Year 4 and Year 3 ($p < .001$). Children in Year 4 were also more accurate than those in Year 3 ($p < .001$). The significant differences between schools, $F(6, 691) = 7.45$, $p < .001$, $\eta_\rho^2 = .06$, were a result of pupils in sch7 performing less accurately than those in sch1, sch2, and sch4 ($p < .003$). Children in sch3 also performed less well than those in sch1 ($p = .017$).

4.3.1.4. English spelling. As with the previous analyses, there were significant main effects of year group, $F(2, 691) = 42.69$, $p < .001$, $\eta_\rho^2 = .11$, and school, $F(6, 691) = 10.26$, $p < .001$, $\eta_\rho^2 = .08$. Children in Year 5 were more accurate than those in Years 3 ($p < .001$) and 4 ($p < .001$). Children in Year 4 were also more accurate than those in Year 3 ($p < .001$). Pupils in sch7 were less accurate spellers than those in sch1, sch2, sch4, and sch6 ($p < .011$). Children in sch3 were also less accurate than children in sch1 ($p < .019$).

These analyses reveal consistent year group effects across Welsh and English literacy tests, where accuracy improved with increasing age. There were also school effects across all Welsh and English tests. Whilst these effects differed according to task, a consistent finding

was that children in the Welsh medium school (sch7) outperformed children from other schools on the Welsh tasks, particularly in spelling. Conversely, performance by these children on English literacy tests was relatively poor, again, particularly in spelling. The atypical profiles presented by children in sch7 are likely to be due to the language background of the school and its pupils, however, seeking the cause of the deflated English literacy scores among pupils in sch7 is beyond the scope of this study. In sum, the current analyses present clear evidence the literacy profiles of children in sch7 are somewhat different to pupils in the other schools meaning any further investigations using participants from sch7 would likely be confounded by language. For this reason, pupils from sch7 were removed from the current and subsequent studies with this sample. It is important to note that there were also some fluctuations in performance between the English medium schools. When considering performance across all four tasks, these fluctuations appear to be random and likely reflect minor variations in the emphasis placed on Welsh language and literacy instruction.

Table 4.3.

Means and Standard Deviations for Each Year Group Across Schools on Welsh and English Literacy Tests

	Sch1	Sch2	Sch3	Sch4	Sch5	Sch6	Sch7	Simple Effects Analyses
					Welsh Literacy			
Reading								
Year 3	26.05 (7.21)	24.66 (6.94)	19.43 (7.37)	24.23 (7.20)	20.56 (7.28)	24.72 (6.92)	29.27 (8.21)	sch3 < sch1**, sch7***
Year 4	26.55 (6.92)	18.52 (7.47)	25.00 (8.32)	27.96 (5.28)	23.75 (9.57)	27.94 (5.76)	33.03 (9.07)	sch7 > sch1*, sch3***, sch5*** sch2 < sch1**, sch6**
Year 5	28.86 (4.73)	26.62 (9.25)	25.08 (7.42)	26.15 (7.14)	26.28 (7.41	32.00 (8.48)	34.68 (7.53)	sch7 > sch2*, sch3***, sch4*
Spelling								sch7 > sch1 – sch6***
Year 3	15.98 (3.39)	16.30 (2.42)	13.17 (2.51)	18.43 (3.55)	15.89 (3.66)	25.00 (6.05)	31.93 (2.46)	sch6 > sch1 – sch5***
Year 4	20.24 (4.15)	17.86 (3.91)	15.68 (3.82)	20.21 (3.77)	17.31 (4.27)	25.65 (5.89)	34.85 (3.26)	sch7 > sch1 – sch6*** sch6 > sch1 – sch5***
Year 5	19.10 (3.15)	17.72 (4.21)	18.18 (4.05)	22.07 (5.40)	18.61 (4.17)	27.39 (7.37)	34.00 (3.19)	sch7 > sch1 – sch6*** sch6 > sch1 – sch5*
					English Literacy			
Reading								
Year 3	17.80 (4.68)	16.50 (4.31)	13.57 (4.67)	16.67 (4.78)	14.89 (4.79)	16.66 (4.65)	13.96 (6.27)	
Year 4	20.81 (5.73)	19.48 (6.16)	18.73 (5.73)	21.67 (4.67)	20.00 (4.99)	19.67 (5.87)	16.62 (6.19)	sch1 > sch7*
Year 5	22.96 (4.86)	25.07 (6.90)	20.88 (6.51)	22.54 (7.26)	21.15 (7.19)	20.89 (7.10)	19.16 (5.42)	
Spelling								
Year 3	26.98 (4.15)	23.33 (3.75)	23.03 (5.83)	24.43 (4.93)	24.37 (4.99)	24.28 (4.53)	21.64 (5.18)	sch1 > sch7**, sch3*
Year 4	28.81 (5.95)	28.72 (5.08)	26.49 (6.38)	28.52 (5.18)	26.53 (6.09)	26.67 (5.20)	24.56 (5.08)	sch1 > sch7**
Year 5	31.31 (4.59)	29.93 (4.88)	30.03 (5.58)	30.32 (6.73)	28.85 (5.84)	29.85 (5.37)	26.10 (4.32)	sch1 > sch7**

Note. Standard deviations are reported in parentheses next to means.
[a]Simple effects analyses were run on estimated marginal means of each school within year group using Bonferroni corrections.

3.3.2. Developmental Sensitivity of Screening Measures

After removing children from sch7, a second set of analyses were run to ensure each screening test was sensitive to age (year group) and to check for any consistent school clustering effects in the tests used for measuring English literacy and motor related skills. To test for year group and school effects, a series of weighted two-way ANOVAs were run on the scores from each test. Significant effects were followed up using pairwise comparisons with Bonferroni corrections.

3.3.2.1. Literacy tests. Table 4.4 reports the descriptive statistics for accuracy of each year group within the schools on the English reading and spelling tests. Accuracy increased with age and was similar across schools. The two-way ANOVAs (see Table 4.5) confirmed that – across all English literacy measures – accuracy increased significantly with age. That is, children in Year 5 were more accurate than those in Year 4 who were more accurate than those in Year 3. There was a small, but consistent, effect of school, due to children in sch1 being more accurate than children in sch3 across all tests.

3.3.2.2. Motor tests. On the whole, performance on motor tests improved with age and was similar across schools (see Table 4.6). Two-way ANOVAs on the Coding and Visual Motor Integration tests showed performance increased significantly with age whereby children in Year Five were more accurate than children in Year 4 who were more accurate than children in Year 3 (see Table 4.7). There was no effect of school on Visual Motor Integration accuracy and no differences between schools on the Coding fluency at follow-up. There was also a significant effect of year group on the Matrices and Handwriting Legibility tests where accuracy/legibility was lower in Year 3 than in Year 4 and Year 5. There were also effects of school on the Matrices accuracy and Handwriting Legibility tests. School differences on the Matrices were due to pupils in sch3 being more accurate than children in sch5. Whereas, differences in Handwriting Legibility were driven by greater legibility in sch1

than in sch2. School differences in handwriting legibility are likely to reflect varying emphases on handwriting instruction between schools (Graham et al., 2008; see Chapter 1).

The ANOVAs were run to assess the age sensitivity and school clustering effects of the battery. On the whole, performance on the screening tests increased with year group. Across all tests, pupils in Years 4 and 5 were more accurate than pupils in Year 3. In the majority of tests, pupils in Year 5 were also more accurate than pupils in Year 4. The analyses also revealed performance was influenced by school on some of the screening tests. However, there was no consistent pattern of school effects across the screening tests and the effect sizes of differences between schools were small, suggesting only minor variations. Thus, on the whole, the battery appears to be sensitive to age but not to school effects. With this in mind, the ensuing analyses examined the relationships between screening measures between year groups but collapsing across schools.

Table 4.4.

Means, Standard Deviations, and Reliabilities of English Literacy Tests Across Age Groups and Schools

	Schools						Reliabilities[a]	
	Sch1	Sch2	Sch3	Sch4	Sch5	Sch6	Published	Reported
Reading								
Year 3	17.80 (4.68)	16.50 (4.31)	13.56 (4.67)	16.67 (4.78)	14.89 (4.79)	16.66 (4.65)	-	.87
Year 4	20.81 (5.73)	19.48 (6.16)	18.73 (5.73)	21.67 (4.67)	20.00 (4.99)	19.67 (5.87)	-	.91
Year 5	23.00 (5.87)	25.07 (6.90)	20.88 (6.51)	22.54 (7.26)	21.15 (7.19)	20.89 (7.10)	-	.92
Word Spelling								
Year 3	26.98 (4.15)	23.33 (3.75)	23.03 (5.83)	24.43 (4.93)	24.37 (4.99)	24.28 (4.53)	.81	.87
Year 4	28.81 (5.95)	28.72 (5.08)	26.49 (6.38)	28.52 (5.18)	26.53 (6.09)	26.67 (5.20)	.86	.90
Year 5	31.39 (5.21)	29.93 (4.88)	30.03 (5.58)	30.32 (6.73)	28.85 (5.84)	29.85 (5.37)	.89	.89
Sentence Spelling								
Year 3	39.47 (9.47)	32.87 (9.85)	29.69 (12.64)	35.57 (12.65)	33.58 (11.05)	36.10 (9.33)	-	.93
Year 4	42.53 (10.06)	41.93 (11.10)	37.90 (12.93)	45.63 (7.31)	40.25 (10.99)	37.69 (10.59)	-	.93
Year 5	45.49 (9.64)	46.53 (6.78)	45.30 (9.80)	46.11 (9.70)	44.50 (10.58)	43.94 (10.57)	-	.92

Notes. Standard deviations are reported in parentheses. Dashes denote instances where published reliabilities were not available.
[a] Internal consistency (Cronbach's alpha).

Table 4.5.

Two-Way ANOVAs with Post-hoc Comparisons of Year Group and School Differences on

English Literacy Tests

	F	η_ρ^2	Post-Hoc Comparisons
Cloze Reading			
Year Group	41.52***	.12	5 > 4*** > 3***; 4 > 3***
School	4.65***	.04	Sch1 > Sch3*
Word Spelling			
Year Group	38.02***	.11	5 > 4*** > 3***; 4 > 3***
School	4.91***	.04	Sch1 > Sch3*
Sentence Spelling			
Year Group	36.54***	.11	5 > 4*** > 3***; 4 > 3***
School	4.30***	.03	Sch1 > Sch3*

Note. ***$p < .001$, ** $p < .01$, *$p < .05$.

Table 4.6.

Means, Standard Deviations, and Reliabilities of Motor Tests Across Age Groups and Schools

	Schools						Reliabilities	
	Sch1	Sch2	Sch3	Sch4	Sch5	Sch6	Published	Reported
Matrices								
Year 3	26.00 (5.50)	25.47 (5.47)	27.85 (4.81)	25.07 (6.59)	22.79 (5.40)	28.48 (4.90)		.77[a]
Year 4	30.90 (3.30)	31.10 (4.32)	30.38 (5.45)	31.50 (5.66)	28.65 (3.55)	30.15 (3.44)	.93[a]	.78[a]
Year 5	31.28 (3.98)	29.38 (6.99)	33.38 (3.82)	33.96 (4.44)	30.94 (4.70)	29.64 (5.20)		.77[a]
Coding								
Year 3	35.89 (7.59)	32.90 (7.49)	30.80 (7.11)	34.17 (7.71)	30.89 (7.22)	28.86 (5.55)	.83[a]	.91[a]
Year 4	35.29 (8.09)	36.45 (8.21)	34.90 (7.80)	39.71 (6.62)	37.29 (5.22)	33.97 (7.18)	.83[a]	.78 [a]
Year 5	39.30 (8.41)	40.28 (8.93)	37.35 (6.78)	38.93 (8.66)	40.56 (9.70)	38.64 (7.11)	.89[a]	.70 [a]
Visual Motor Integration								
Year 3	19.07 (2.31)	18.67 (2.63)	19.03 (2.20)	19.45 (2.97)	19.00 (2.58)	19.90 (2.88)	.82[a]	.76[a]
Year 4	20.88 (2.71)	20.97 (3.32)	20.10 (3.25)	21.52 (3.13)	20.18 (2.65)	20.56 (2.99)	.79[a]	.76[a]
Year 5	22.63 (2.72)	20.90 (4.34)	21.77 (3.54)	21.89 (3.60)	21.95 (3.50)	21.17 (2.99)	.81[a]	.73[a]
Handwriting Legibility								
Year 3	17.97 (4.12)	15.70 (5.02)	16.97 (4.20)	16.37 (4.85)	17.89 (4.85)	17.79 (3.10)	-	.84[b]
Year 4	21.36 (4.15)	18.79 (5.00)	19.02 (4.22)	19.26 (4.51)	20.81 (2.86)	18.66 (3.90)	-	.86[b]
Year 5	21.61 (3.72)	18.30 (4.42)	20.60 (3.25)	20.32 (3.80)	20.40 (3.94)	19.86 (3.50)	-	.77[b]

Notes. Dashes denote instances where published reliabilities were not available.
[a]Internal consistency (Cronbach's alpha). [b]Inter-rater (two-way random effects intra-class correlation).

Table 4.7.

Two-Way ANOVAs with Post-hoc Comparisons of Year Group and School Differences on

Motor Tests

	F	η_{ρ}^2	Post-Hoc Comparisons
Matrices			
Year Group	47.78***	.14	5 > 3***; 4 > 3***
School	2.71*	.02	Sch3 > Sch5**
Coding			
Year Group	56.53***	.08	5 > 4*** > 3***; 4 > 3***
School	3.56**	.03	-
Visual Motor Integration			
Year Group	25.15***	.08	5 > 4* > 3***; 4 > 3***
School	1.13	.01	-
Handwriting Legibility			
Year Group	21.25***	.07	5 > 3***; 4 > 3***
School	6.24***	.05	Sch1 > Sch2***

Note. ***$p < .001$, ** $p < .01$, *$p < .05$.

4.3.3. Relationships Within and Between Literacy and Motor Measures

Pearson's correlations were run for each year group across all tests to assess the

strength of relationships between measures (see Tables 4.8 – 4.10). It was expected that

measures tapping similar constructs (literacy or motor) would correlate well, thus

demonstrating convergent validity. Weak correlations between measures hypothesised to tap

different constructs would demonstrate divergent validity.

4.3.3.1. Literacy tests. Large correlations were found between all three English

literacy measures – Cloze Reading, Word Spelling, and Sentence Spelling – particularly

between the spelling tests. The strength and pattern of these correlations was consistent

across all three-year groups, indicating the reading and spelling tests had convergent validity

and were consistent measures of English literacy.

4.3.3.2. Motor tests. Varying relationships were found amongst Matrices, Coding,

Visual Motor Integration and Handwriting Legibility tests. Significant correlations were

found between published tests purporting to measure aspects of visuospatial and/or

graphomotor skills. Specifically, the Matrices test correlated with the Visual Motor Integration test, producing similar medium sized correlations in all year groups and demonstrating convergent validity as both tasks tap visuospatial skills (Prunty et al., 2016). To a lesser extent, both Matrices and Visual Motor Integration tests correlated with the Coding test – a measure of graphomotor speed – suggesting Coding also shares variance with visual perceptual skills (see Tables 4.8 – 4.10).

Medium to large correlations were found across year groups on Handwriting Legibility dimensions, Letter Formation, Letter Spacing, Word Spacing, and Line Alignment (see Tables 4.8 – 4.10) indicating convergent validity. In particular, there were strong associations between Letter Formation and Letter Spacing which were expected as both dimensions assess legibility at the letter level. Another strong relationship was found between Letter and Word Spacing in Years 3, 4, and – to a lesser extent – Year 5 which was to be expected as both dimensions were measuring spatial features of handwriting. However, the Script Switching dimension did not correlate well with any other dimension, and, more generally, failed to correlate with any other test, suggesting that this dimension may be capturing an alternate underlying construct.

Finally, correlations between the published tests of visuospatial and graphomotor skills with the Handwriting Legibility dimensions revealed varying relationships between the measures. The Visual Motor Integration test and Legibility dimensions, both of which require skills related to pencil control, yielded small to medium correlations between (see Tables 4.8 – 4.10), suggesting that these measures tap somewhat different aspects of motor skills. There were weak relationships between the Legibility dimensions and Matrices and Coding tests. The poor relationship between legibility and the Matrices also suggests that these measures tap different aspects of visuospatial skills. Similarly, the weak relationship between Handwriting Legibility and Coding – despite both tasks tapping graphomotor skills – is likely

to be due to different task demands whereby the Coding test measured graphomotor speed (fluency), the Handwriting Legibility test measured accuracy. Therefore, whilst the Matrices, Coding, and Visual Motor Integration tests likely tap visuospatial and motor coordination considered as perceptuomotor skills the Handwriting Legibility dimensions appear to measure more specific motor action.

4.3.3.3. Literacy and motor tests. Generally, small to medium correlations were found between the literacy and motor tests indicating divergent validity (Tables 4.8 – 4.10). However, significant relationships between tests measuring these two constructs were found. These associations could be explained by shared task demands beyond the construct the test was primarily measuring. For example, small to medium correlations between the Matrices and English literacy tasks (see Tables 4.8 – 4.10) presumably reflect general ability. The moderate correlations found between the English literacy and Visual Motor Integration tests similarly suggest some shared task variance. For example, these measures share the need for pencil manipulation skills. Furthermore, correlations between literacy and the Coding tests may be due to shared speed of processing task demands.

Medium correlations between the English spelling tests and letter formation ratings (see Tables 4.8 – 4.10) were also found, which suggest a relationship between these skills. In turn, the smaller correlations across all years between the spelling tasks and the letter spacing ratings point to the specificity of the spelling-letter formation relationship.

Table 4.8.

Correlations Among Screening Measures of Children in Year Three

	1	2	3	4	5	6	7	8	9	10	11	12
1. Matrices	-											
2. Coding	.26***	-										
3. Word Spelling	.24***	.42***	-									
4. Sentence Spelling	.31***	.41***	.87***	-								
5. Reading	.31***	.41***	.67***	.67***	-							
6. HW - Letter Formation	.13	.26***	.43***	.43***	.26***	-						
7. HW - Letter Spacing	.16*	.22**	.23**	.21**	.10	.64***	-					
8. HW - Word Spacing	.17*	.15*	.20**	.21**	.11	.35***	.52***	-				
9. HW - Line Alignment	.03	.09	.18*	.13	.06	.44***	.43***	.20**	-			
10. HW - Script Switching	.01	-.15	-.13	-.10	-.08	-.03	-.03	-.10	.11	-		
11. HW - Total	.16*	.17*	.27***	.27***	.14	.70***	.79***	.66***	.66***	.31***	-	
12. VMI	.41***	.26***	.25***	.29***	.22**	.18**	.18*	.20**	-.00	-.05	.18**	-

Note. N = 208. HW = Handwriting. MI = Visual Motor Integration
*p < .05, ** p < .01, ***p < .001

Table 4.9.

Correlations Among Screening Measures of Children in Year Four

	1	2	3	4	5	6	7	8	9	10	11	12
1. Matrices	-											
2. Coding	.19**	-										
3. Word Spelling	.36***	.22**	-									
4. Sentence Spelling	.40***	.30***	.87***	-								
5. Reading	.34***	.30***	.69***	.73***	-							
6. HW - Letter Formation	.19**	.13	.33***	.40***	.22**	-						
7. HW - Letter Spacing	.25***	.19**	.23***	.28***	.18**	.62***	-					
8. HW - Word Spacing	.17*	.14*	.11	.17*	.14*	.43***	.54***	-				
9. HW - Line Alignment	.12	.05	.20**	.20**	.10	.48***	.50***	.29***	-			
10. HW - Script Switching	-.06	-.15*	-.02	-.04	-.07	-.09	-.02	-.07	-.02	-		
11. HW - Total	.19**	.09	.27***	.32***	.20**	.73***	.80***	.70***	.65***	.29***	-	
12. VMI	.40***	.20**	.36***	.44***	.32***	.33***	.29***	.20**	.22**	-.02	.31**	-

Note. N = 212. HW = Handwriting. VMI = Visual Motor Integration
*p < .05, ** p < .01, ***p < .001

Table 4.10.

Correlations Among Screening Measures of Children in Year Five

	1	2	3	4	5	6	7	8	9	10	11	12
1. Matrices	-											
2. Coding	.28***	-										
3. Word Spelling	.24***	.26***	-									
4. Sentence Spelling	.29***	.28***	.82***	-								
5. Reading	.29***	.37***	.67***	.73***	-							
6. HW - Letter Formation	.23**	.16*	.34***	.40***	.20**	-						
7. HW - Letter Spacing	.06	.11	.22**	.19**	.05	.62***	-					
8. HW - Word Spacing	-.03	.14	.05	.04	-.04	.30***	.38***	-				
9. HW - Line Alignment	.10	.14	.23**	.19**	.13	.49***	.46***	.27***	-			
10. HW - Script Switching	.22**	-.08	.05	.06	.04	.10	.02	-.08	.05	-		
11. HW - Total	.19**	.14*	.26***	.26***	.10	.75***	.75***	.61***	.67***	.38***	-	
12. VMI	.36***	.22**	.43***	.38***	.30***	.30***	.24***	.12	.20**	.03	.28***	-

Note. N = 202. HW = Handwriting. VMI = Visual Motor Integration
*p < .05, ** p < .01, ***p < .001

4.3.4. Factor Structure of the Screening Battery

Factor analysis techniques were used to confirm the hypotheses of screening tests loading onto distinct literacy, perceptuomotor, and handwriting factors and to examine the underlying factor structure of the battery in detail. Doing so is necessary to validate the battery and to identify the measures which have the greatest loadings onto their factors.

4.3.4.1. Exploratory factor analysis. Prior to running confirmatory factor analyses (CFA), exploratory factor analyses (EFA) were run across the whole sample to validate the theoretically driven three-factor solution rather than a two- or four-factor solution. All measures were used in the analysis apart from the Script Switching and the Total Handwriting Legibility Dimension as well as the Welsh Literacy measures. The Script Switching measure was removed as it correlated poorly with all other variables and the Handwriting Total Legibility measure was not included as it was a composite of all other Handwriting Legibility measures. Welsh Literacy Measures were excluded as the primary interest in this study was English literacy proficiency.

Exploratory factor analysis with Maximum likelihood (ML) estimation and geomin rotation was used in Mplus 7.2 (Muthén & Muthén, 2014). ML estimation was used as it provides goodness-of-fit indices to compare the two, three, and four factor solutions. Furthermore, correlations between factors were expected (see previous section), thus geomin (oblique) rotation was used rather than an orthogonal method.

Model fit indices and difference test between models are reported in Table 4.11. The guidelines proposed by Hu and Bentler (1999) were used to assess model fit in this and subsequent analyses in later studies. The guidelines suggest a well-fitting model should have as a minimum: χ^2 with $p > .05$, RMSEA $< .06$, SRMR $< .08$, CFI and TLI $> .95$. Based on these criteria the two-factor solution was a poor fit, whilst both three and four factor solutions provided a satisfactory fit to the data.

Table 4.11.

Goodness-of-Fit and Chi Square Difference Tests for Two, Three, and Four Factor

Exploratory Factor Analyses of Screening Battery Measures

Solution	χ^2	df	RMSEA	SRMR	CFI	TLI	χ^2_{diff}	Δ_{df}
2-Factor	123.44***	26	.08	.04	0.96	.93	-	-
3-Factor	44.38***	18	.05	.02	0.99	.97	79.05***[a]	8
4-Factor	22.95*	11	.04	.01	1.00	.98	21.43**[b]	7

Note. $N = 626$.
[a]Chi Square difference between two and three factor solutions. [b]Chi Square difference between three and four factor solutions. RMSEA, root mean square of error approximation; SRMR, standardised root mean square residual; CFI, comparative fit index; TLI, Tucker-Lewis index.
*$p < .05$, ** $p < .01$, ***$p < .001$

Examination of the factor loadings on the three- and four-factor solutions reveal good loadings in both models (see Tables 4.12 and 4.13), whereby only Coding had a loading of less than .3 on the three-factor solution, however this measure cross loaded onto two factors in both models. Similarly, communality was good (> .3 for most indicators) in both solutions suggesting measures shared variance.

The pattern of factor loadings in the three-factor solution suggested a more theoretically valid model. For instance, Matrix, Visual Motor Integration, and Coding load onto a single factor, as expected, in the three- but not four-factor model. Theoretical justification for the three-factor solution was corroborated when using the Scree test (Cattell, 1966). Inspection of the eigenvalues for several solutions in Figure 4.2 shows the last substantial decline in eigenvalue was immediately prior to the three-factor solution, indicating the three-factor solution was the most suitable.

Table 4.12

Factor Loadings and Communality Values for the Three-Factor Exploratory Factor Analysis

of the Screening Battery Measures

	Factors			Communality
	1	2	3	
Matrix Reasoning	**.73***	.00	-.12	.48
Coding	**.28***	**.22***	.02	.19
Visual Motor	**.49***	.12	.05	.33
Word Spell	-.06	**.93***	.02	.82
Sentence Spell	.00	**.95***	.00	.90
Reading	.12*	**.72***	-.14*	.59
Letter Form	.00	**.26***	**.67***	.61
Letter Spacing	.09	.00	**.81***	.72
Word Spacing	.13	-.05	**.49***	.29
Line Alignment	-.04	.09	**.56***	.32

Note. Factor loadings > .2 are in boldface.
*p < .05.

Table 4.13

Factor Loadings and Communality Values for the Four-Factor Exploratory Factor Analysis

of the Screening Battery Measures

	Factors				Communality
	1	2	3	4	
Matrix reasoning	.00	**.59**	-.03	.12	.40
Coding	.18	.12	.12*	**.34**	.27
Visual motor	.02	**.65***	.04	-.05	.43
Word spell	**.89***	-.00	.03	.00	.82
Sentence spell	**.92***	.06	.02	-.00	.90
Reading	**.70***	-.02	-.04	**.33***	.67
Letter form	**.23***	.02	**.68***	-.10	.62
Letter spacing	-.03	-.01	**.87***	.06	.73
Word spacing	-.07	.03	**.54***	.12	.30
Line alignment	.06	-.05	**.56***	-.05	.32

Note. Factor loadings > .2 are in boldface.
*p < .05.
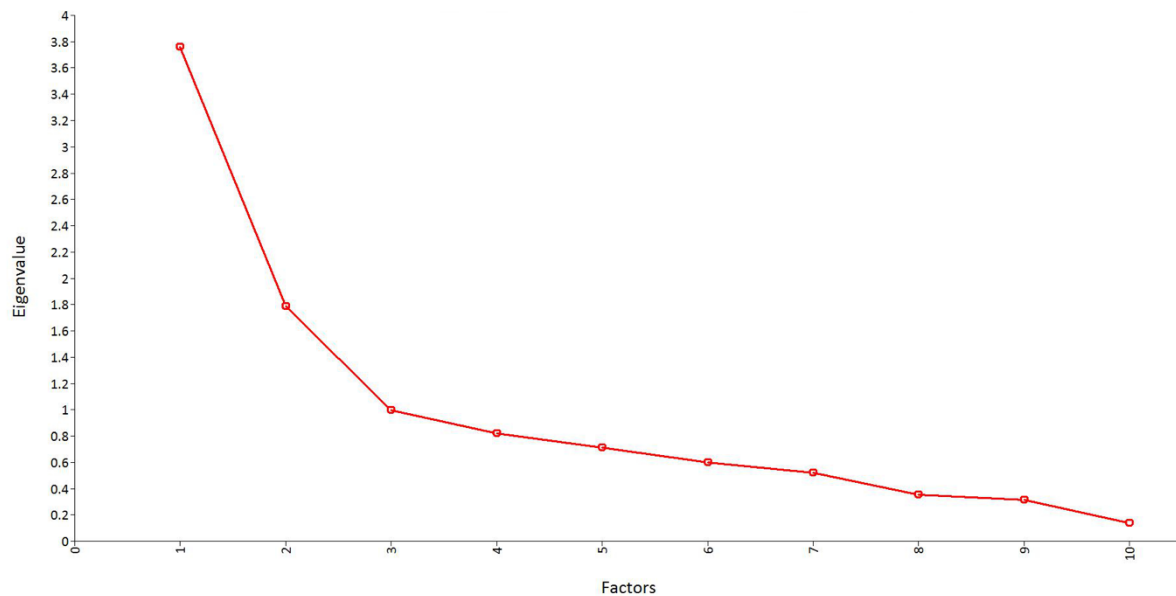
Figure 4.2. A scree plot with Eigenvalues plotted against EFA solutions (one to ten factors), indicating a three-factor solution was optimal.

4.3.4.2. Confirmatory factor analysis. Confirmatory factor analyses (CFA) were used to examine the underlying factor structure of the screening battery, thus empirically validating the tasks which measure a literacy and/or motor skill. Secondly, CFAs were used to identify the measures that had the best factor loadings. Measures with the best factor loadings onto their respective constructs were used as 'marker' measures for identifying children with literacy and/or motor difficulties. Models were initially run on each year group separately (single-group solutions) to identify the best fitting solution for each year group. The best fitting solution was then applied as a multi-group confirmatory factor analysis (MGCFA). The final factor structure of the three factors, literacy, perceptuomotor, and handwriting are presented in Figure 4.3.

4.3.4.2.1. Single-group CFAs. CFAs were run using direct ML estimation in Mplus 7.2 (Muthén & Muthén, 2014) on each year group separately. Direct ML was favoured given a small amount of missing data completely at random (3% in year three and four, 2% in year five). Initially, all measures were entered into the baseline models, only loading onto their

primary hypothesised factors (see Table 4.2). The items of the Visual Motor Integration test were split to create two indicators whilst constraining the factor loadings and residuals between the two. This increased the number of indicators on the perceptuomotor factor and improved the overall model fit.

In two subsequent iterations of the model development, Coding and Letter Formation were freely estimated to cross-load onto the literacy factor. Coding was cross loaded onto the literacy factor as the task shares variance with the literacy tasks via speed of processing. Letter Formation was also cross loaded onto the literacy factor because this measure was likely to be tapping spelling-related letter knowledge as well as motor aspects of letter formation whereas the other handwriting indicators loaded onto the handwriting factor were predominantly measuring motor aspects. The final models were a good fit (see Table 4.14) with no large modification indices, suggesting no areas of strain. All indicators loaded significantly onto their factors, except for the Coding indicator in the Year 5 model which was borderline non-significant ($p = .05$) when loading onto the perceptuomotor factor. The cross-loading was kept in this model as it improved the model fit (see Appendix B for results from separate single-group CFAs).

3.3.4.2.2. Multi-group CFA (MGCFA). The single-group model was then applied to a MGCFA, which allows the testing of measurement invariance to assess whether the measures were comparable across the groups. That is, it ensures the model and the indicators were valid measures across all year groups (Brown, 2015; Milfont & Fischer, 2010). To assess measurement invariance a stepwise procedure of successively more restricted models was used as recommended by Brown (2015), Milfont and Fischer (2010), and Vandenberg and Lance (2000). Using this procedure, the more constrained model is a nested version of the previous model. As such, the new model's goodness-of-fit was examined against the previous, less constrained model. A direct test of fit between the models was completed using

a chi-square difference test to ensure the models do not significantly differ from one another. In addition, a decrease in the CFI magnitude would indicate that the more constrained model be rejected (Cheung & Rensvold, 2002).

Table 4.14 shows that constraining the factor structure yielded configural invariance equal across groups and produced an acceptable fit. This model acted as the baseline for the further, more constrained models. In the next analysis, the equality of factor loadings (metric invariance) was tested between year groups by constraining factor loadings to be equal across groups. Constraining factor loadings gave an overall acceptable model fit. Furthermore, the model did not significantly differ from the configural model, $\chi^2_{diff}(18) = 15.58$, *ns*, nor was there any change in the CFI value. In the next model, the intercepts of the indicators were constrained to be equal across all year groups (scalar invariance). This model did not differ from the less constrained metric invariance model, $\chi^2_{diff}(16) = 0.18$, *ns*.

Invariance across indicator residuals (differences between measurement error between groups) was not tested as it was deemed to be overly restrictive given there were no theoretical or methodological reasons to expect errors to be equal across year groups (see Brown, 2015). Similarly, structural invariance was not tested as performance on indicators was likely to change developmentally (see earlier section on developmental sensitivity). The analyses demonstrate the current solution has measurement invariance, indicating the screening measures were suitable indicators of performance across all year groups. The path diagram of the final model the accompanying unstandardized and standardised factor loadings and indicator residual variances for each year group are presented in Figure 4.3 and Table 4.15.

All indicators significantly loaded onto their respective factors (see Table 4.15) and Visual Motor Integration, Sentence Spelling, and Letter Spacing had the largest factor loadings onto their respective factors across all year groups. There were medium-to-large

correlations were present between Perceptuomotor and Literacy factors (path 14 $r = .42, .58,$ and .54 for Years 3, 4, and 5 respectively), small to medium correlations were present between Literacy and Handwriting factors (path 15 $r = .30, .31,$ and .25 for Years 3, 4, and 5 respectively), and medium correlations were found between Perceptuomotor and Handwriting factors (path 16 $r = .30, .42,$ and .38 for Years 3, 4, and 5 respectively).

Table 4.14.

Goodness-of-Fit Estimates for Single- and Multi-Group Confirmatory Factor Analyses Models of Measures Loading Perceptuomotor, Literacy, and Handwriting

	χ^2	df	RMSEA [90% CI]	SRMR	CFI	TLI	χ^2_{diff}	Δ_{df}
Single-group solutions								
Year 3 ($n = 208$)	$50.03^{n.s.}$	41	.033 [.000, .060]	.044	.99	.99	-	-
Year 4 ($n = 214$)	$50.18^{n.s.}$	41	.032 [.000, .060]	.036	.99	.99	-	-
Year 5 ($n = 204$)	64.39^*	41	.053 [.026, .077]	.048	.97	.96	-	-
Multi-group solutions								
Full configural invariance	173.68^*	125	.043 [.026, .058]	.044	.98	.98	-	-
Full metric invariance	189.26^{**}	143	.039 [.022, .054]	.050	.98	.98	15.58	18
Full scalar invariance	$189.43^{n.s.}$	159	.030 [.000, .046]	.050	.99	.99	0.18	16

Note. $N = 626$. = nested difference between the restricted solution and the preceding less-restricted solution. RMSEA = root mean square of error approximation. 90% CI = 90% confidence intervals for RMSEA. SRMR = standardised root mean square residual. CFI = comparative fit index. TLI = Tucker-Lewis index.
*$p < .05$, ** $p < .01$, ***$p < .001$.
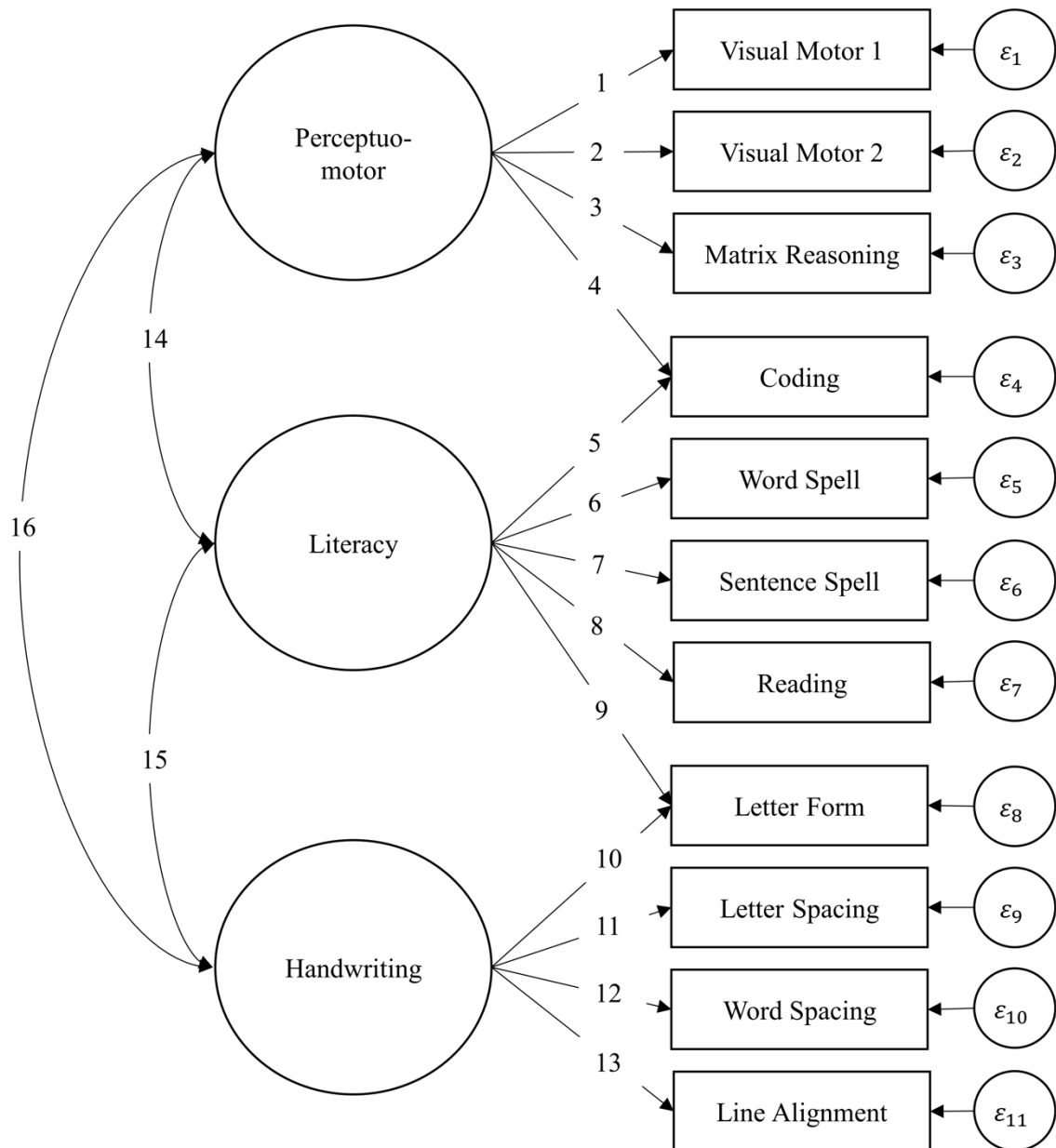
Figure 4.3. Path model of multi-group confirmatory factor analysis (MGCFA) which examined the factor structure of the class screening battery across three-year groups. In each model, 11 measures of literacy and motor skills were loaded onto three factors. Path numbers (1 – 13) correspond to standardised path estimates with residual variances in Table 4.12.

Visual Motor = Visual Motor Integration

All factor loadings for all year groups were significant at $p < .001$.

Table 4.15.

Unstandardised and S*tandardised Factor Loadings of Each Year Group in the Multi-Group Factor Analysis*

| | Unstandardised | | Standardised | | | | | |
| | | | Year 3 | | Year 4 | | Year 5 | |
Path	Estimate	Residual	Estimate	Residual	Estimate	Residual	Estimate	Residual
Perceptuomotor								
1 Visual Motor Integration 1	1.00 (.00)	.45 (.03)	.74 (.03)	.45 (.04)	.74 (.03)	.46 (.04)	.75 (.03)	.55 (.04)
2 Visual Motor Integration 2	1.00 (.00)	.45 (.03)	.74 (.03)	.45 (.04)	.74 (.03)	.46 (.04)	.75 (.03)	.55 (.04)
3 Matrices	.73 (.06)	.71 (.08)	.54 (.05)	.71 (.05)	.54 (.05)	.71 (.05)	.54 (.05)	.29 (.05)
4 Coding	.26 (.07)		.20 (.06)		.19 (.05)		.20 (.05)	
Literacy		.77 (.08)		.83 (.04)		.83 (.03)		.17 (.04)
5 Coding	.30 (.06)		.28 (.05)		.27 (.05)		.26 (.05)	
6 Word Spelling	1.00 (.00)	.16 (.03)	.91 (.02)	.17 (.03)	.90 (.02)	.19 (.03)	.88 (.02)	.23 (.03)
7 Sentence Spelling	1.07 (.03)	.11 (.03)	.95 (.01)	.10 (.03)	.98 (.01)	.05 (.03)	.95 (.02)	.11 (.03)
8 Reading	.83 (.04)	.48 (.05)	.73 (.03)	.46 (.04)	.75 (.03)	.43 (.04)	.76 (.03)	.43 (.04)
9 Letter Formation	.25 (.04)		.23 (.04)		.22 (.03)		.23 (.04)	
Handwriting		.40 (.05)		.40 (.05)		.41 (.05)		.41 (.05)
10 Letter Formation	1.00 (.00)		.68 (.04)		.67 (.04)		.68 (.04)	
11 Letter Spacing	1.26 (.09)	.24 (.06)	.87 (.04)	.25 (.06)	.87 (.03)	.25 (.06)	.83 (.04)	.75 (.06)
12 Word Spacing	.79 (.07)	.70 (.07)	.54 (.04)	.70 (.05)	.56 (.04)	.68 (.05)	.51 (.04)	.30 (.05)
13 Line Alignment	.83 (.07)	.73 (.08)	.55 (.04)	.70 (.05)	.58 (.04)	.67 (.05)	.56 (.04)	.30 (.04)

Note. Path numbers correspond to those presented in the path diagram (Figure 4.2). Residuals correspond to the standardised indicator residual variances. Standard errors are reported in parentheses next to the loading/residual.
All factor loadings *p* < .001.

4.3.5. Prevalence of Literacy and Motor Difficulties

Measures used to identify children as having literacy, motor, or comorbid literacy and motor difficulties were selected based on their relationships with other measures (correlations), their ability to measure the construct of interest (factor loadings) and judgement. The measures selected to identify literacy difficulties were the Cloze Reading, Word Spelling, and Sentence Spelling tests. All three measures had good reliabilities, correlated well with one another, and had excellent factor loadings.

The measures selected to identify motor difficulties were the Visual Motor Integration test, Letter Formation dimension, and the overall Handwriting Legibility Score. The Visual Motor Integration test was selected owing to its good factor loadings on the perceptuomotor factor. Furthermore, it was less likely to be confounded by non-verbal ability than the other perceptuomotor measures. The Letter Formation dimension also had good factor loadings onto the handwriting factor and was selected over the Letter Spacing measure – which had better factor loadings on the same construct – as both had excellent correlations with one-another but the Letter Formation scale had a slightly stronger relationship with the Visual Motor Integration test across the year groups. It was also deemed more likely to capture motor execution processes (e.g., fine motor skill) than the Letter Spacing measure which presumably captured more spatial planning processes. Finally, the total Handwriting Legibility Score was selected, despite not being included in the CFA, to act as a holistic measure of Handwriting given that it correlated well with the Handwriting Legibility dimensions which all loaded onto the construct well. Using Visual Motor Integration test fulfilled Criterion A (coordinated motor skills below the expected standard) whilst two handwriting measures also fulfilled criterion B (difficulties interfere with daily activities) of DCD in the DSM-V (APA, 2013).

A marker approach (see Snowling & Hulme, 2015) was used to identify children with literacy and motor difficulties. Children who scored below the cut-off, of < 1.33 *SD* below their year group average on two out of three of the selected literacy tests – Cloze Reading, Word Spelling, or Sentence Spelling – only were identified as having literacy difficulties. Children who scored below the cut-off on two out of the three selected motor measures – Visual Motor Integration, Letter Formation, and Handwriting Legibility – only were identified as having motor difficulties. Children who met the criteria for both literacy and motor difficulties were identified as having comorbid literacy and motor difficulties. Children who did not meet any criteria were labelled as typically developing (TD).

The numbers of children identified as having literacy, motor, co-morbid literacy and motor difficulties, or being typically developing are reported in Table 4.16. The percentage of children with a literacy-only, motor-only, and comorbid literacy and motor difficulties or who were typically developing was calculated by dividing the number of children in the group by the total number of children in the sample. These percentage estimates are reported alongside previously published population estimates of literacy and/or motor difficulties. The published population estimates for singular disorders were taken from studies examining the prevalence of the specific disorder in large community-based samples in the UK. The published population estimate for comorbid literacy and motor difficulties was taken from Cruddace and Riddell (2006) data as the only study to assess the prevalence of literacy and motor difficulties in a community sample.

Table 4.16.

Proportion of Children Identified with Literacy, Movement, Comorbid Difficulties, or

Typically Developing Compared Against Published Estimates

	n	%	Published estimates (%)
Literacy difficulties/dyslexia	**42**	**7**	
Year 3	11	6	10.5[b]
Year 4	17	8	
Year 5	14	7	
Motor difficulties/DCD	**34**	**6**	
Year 3	12	6	4.9[c]
Year 4	8	4	
Year 5	14	8	
Comorbid literacy and motor difficulties (dyslexia and DCD)[a]	**16**	**3**	
Year 3	6	3	13[d]
Year 4	6	3	
Year 5	4	2	
Typically developing	**513**	**84**	
Year 3	170	85	-
Year 4	176	85	
Year 5	167	83	

Note. Subtotal sample breakdown per group are reported in boldface.
[a]Percentage of children with comorbid difficulties relative to children who meet the criteria for at least one disorder. [b](Snowling and Hulme, 2015). [c](Lingham et al., 2009). [d](Cruddace & Riddell, 2006).

The derived prevalence estimates were examined to determine whether the frequency of comorbid literacy and motor difficulties exceeds that expected by chance. To do so, the percentage of expected cases of comorbid literacy and motor difficulties was calculated by multiplying the (base) rates of singular literacy and motor difficulties. The expected rate ($n = 3$, 0.54%) was then compared with the number of observed cases ($n = 16$; 3.31%) as described by Caron and Rutter (1991) and Landerl and Moll (2010). The observed frequency of children with comorbid literacy and motor difficulties was significantly higher than those

expected by chance (OR = 5.78, p < .001), suggesting comorbid literacy and motor difficulties cannot be attributed to chance alone.

4.4. Discussion

Prior to investigating impairments in transcription processes amongst children with dyslexia, DCD, and comorbid dyslexia and DCD, it was first necessary to identify children at risk of these disorders and to examine their prevalence in a large community-based sample. In this study, we identified similar rates of singular literacy and motor difficulties to those reported in the literature. The prevalence of comorbid literacy and motor difficulties was smaller than has previously been reported, but the prevalence of comorbid difficulties was still greater than would be expected if there was no relationship between the disorders. In the rest of this discussion, we discuss this investigation's prevalence rates in relation to the wider literature and its implications.

4.4.1. Prevalence of Singular Literacy and Motor Difficulties

Despite differences in how literacy and motor difficulties were operationalised and assessed, the prevalence rates of singular literacy and motor difficulties found here corroborate estimates of dyslexia and DCD widely reported in the literature (see Blank et al, 2012; Snowling, 2013). Good agreement between the current prevalence rates and those reported in the literature suggest the current rates are accurate and validates the class screening/marker approach in identifying children who have literacy and/or motor difficulties (at risk of dyslexia and/or DCD). Accurate prevalence estimates of singular disorders drawn from the general population are crucial for investigating comorbid disorders because they act as base rates in establishing whether the prevalence of comorbid disorders is greater than would be expected by chance (Caron & Rutter, 1991). Prior to discussing the prevalence of comorbid difficulties, the prevalence of singular literacy and motor difficulties found in this study are compared with previous studies estimating either dyslexia or DCD.

4.4.1.1. Literacy Difficulties. Of the children screened in this study, 7% had literacy difficulties, or, were at risk of dyslexia. This is within the widely reported 3 – 10% prevalence rate of dyslexia (Shaywitz et al., 1990; Snowling, 2013) but below that of the most recent UK estimate of 10.5% reported by Snowling and Hulme (2015). The small difference in prevalence estimates between the current study and that reported by Snowling and Hulme (2015) is counterintuitive given that less conservative cut-offs were used here. However, the difference in prevalence rates could be attributed to differences in how we operationalised and measured literacy difficulties.

Children with literacy difficulties were identified based on their performance on both reading and spelling tests, as opposed to previous investigations which have focused exclusively on reading (Rutter et al., 2004; Shaywitz et al., 1990, Snowling & Hulme, 2015). Children were classified on spelling ability in this study because spelling difficulties feature in current definitions of dyslexia (Rose, 2009) and are more resistant to remediation than reading difficulties (Romani et al., 2005). It is important to note here that some investigations have found reading and spelling skills to be dissociable amongst German-speaking children (Moll & Landerl, 2009). However, we found reading and spelling skills to share a large amount of variance and therefore were highly related skills with no evidence of dissociation in English speaking children.

As a consequence of measuring both reading and spelling skills to identify literacy difficulties, children completed several tests on two separate occasions. This was a broader approach than previous investigations that have focused exclusively on reading ability alone (Rutter et al., 2004; Shaywitz et al., 1990; Snowling et al. 2009). The benefit of administering multiple measures over two sessions is that it reduces the risk of false positives (i.e., wrongly categorising a child as being at risk of dyslexia when they were not). This more conservative approach could explain why the prevalence of literacy difficulties was smaller than those

reported by Snowling and Hulme (2015). In sum, our estimates of literacy difficulties fall within the prevalence estimates widely reported in the literature. However, our rates were slightly below those reported by the most recent epidemiological survey of dyslexia in the UK (Snowling & Hulme, 2015) and possibly reflects a more conservative approach we used in identifying literacy difficulties.

4.4.1.2. Motor Difficulties. We identified 6% of the sample as having motor difficulties, or, being at risk of DCD. The current prevalence rate corroborates widely reported 'ball park' prevalence figures (Blank et al., 2012) and the estimate of 4.9% from a UK based epidemiological investigation (Lingam et al., 2009). The corroboration between our prevalence estimates and the rate reported by (Lingam et al., 2009) is of particular interest due to differences in how motor difficulties are defined and operationalised in the literature. To mitigate the issues associated with using common motor skill tests to identify DCD (Mcintyre et al., 2017; Smits-Engelsman et al., 2015), perceptuomotor and handwriting tests which tap motor processes were used to assess motor skills. As such, a related aim was to assess whether the measures we used to identify children at risk of DCD were suitable for doing so. Therefore then, the high corroboration between Lingam et al. (2009) – who identified children with DCD using rigorous criteria which strictly followed a clinical diagnostic protocol – and our own prevalence rates adds credibility to the validity of our alternate approach to identifying children at risk of DCD.

4.4.2. Prevalence of Comorbid Literacy and Motor Difficulties

The frequency of children with comorbid literacy and motor difficulties found in this study was lower than those reported in previous community (Cruddace & Riddell, 2006) and clinic (Kaplan et al., 1998) samples. Specifically, 3% of the entire sample studied here had comorbid literacy and motor difficulties whereas Cruddace and Riddell (2006) – who also assessed children in community primary schools – identified 13% of children with comorbid

reading and motor difficulties. The authors reported relatively high prevalence rates of singular reading and motor difficulties suggesting their sample was not representative of the general population. Indeed, the class teachers in the study noted there was an unexpected number of children with developmental disorders in the classes that were tested. The abnormally high rates of developmental disorders in Cruddace and Riddell's (2006) sample may explain why their estimates of comorbid difficulties were larger than the ones we found in this investigation. An alternative interpretation of the lower rates found in this study when compared with Cruddace and Riddell (2006) is that it reflects the relatively restricted range of motor tests (e.g., no measure of gross motor skill) used in the current study. However, the similarity in the frequency estimates between our study and Lingham et al. (2009) suggests this latter explanation is unlikely.

To make the current prevalence rates comparable to Kaplan et al.'s (1998) clinic based estimates it is necessary to compare the number of children with comorbid literacy and motor difficulties with the number of children who met the criteria for either literacy and/or motor difficulties. Using the adjusted estimates, 17% of children in the current sample had comorbid literacy and motor difficulties which was below the 33% prevalence rate reported by Kaplan et al. (1998). The high frequency of comorbid dyslexia and DCD reported by Kaplan et al. (1998) relative to our estimates reflects the clinic sampling used by Kaplan et al. (1998). Clinic samples include a disproportionately high number of children with comorbid dyslexia and DCD because the likelihood of referral for these individuals increases as a function of the combined likelihood of referral for dyslexia and DCD separately. This increased referral likelihood is also compounded by referral biases practices (Caron & Rutter, 1991). Therefore, the differences in prevalence rates of comorbid literacy and motor difficulties between the current study and that by Kaplan et al. (1998) reflects variations in sampling methods.

Although there is little agreement in the exact prevalence rates of comorbid literacy and motor difficulties, regardless of sampling differences all investigations have reported a disproportionately high frequency of comorbid disorders (Cruddace & Riddell, 2006; Kaplan et al., 1998; Schoemaker et al., 2013). This corroborates the current findings where the frequency of children with comorbid literacy and motor difficulties was greater than would be expected by chance alone.

4.4.3. Implications for Understanding Comorbidity

The high frequency of comorbid literacy and motor difficulties reported here indicates that literacy and motor disorders are not completely independent but are related to one another. Multifactorial, but not single deficit theories, can account for the increased frequency of comorbidity found here (see Chapter 2). Indeed, comorbid literacy and motor difficulties can be conceptualised in Pennington's (2006) multiple deficit model (MDM). According to this model, dyslexia (literacy difficulties) and DCD (motor difficulties) are a result of independent and shared aetiological and cognitive risk factors. Therefore, children with shared aetiological and cognitive risk factors of dyslexia and DCD are likely to have comorbid literacy and motor difficulties.

Establishing an increased prevalence of comorbid literacy and motor difficulties than was expected based on the rates of singular disorders suggests an underlying relationship between literacy and motor difficulties, possibly due to shared aetiological and cognitive risk factors. However, it is important to note that in this study we assessed motor skills most related to fine motor skills and not global motor skills. Furthermore, the exact relationship between literacy and motor difficulties remains unclear and the high frequency of children with comorbid difficulties found in our sample could be explained by several competing hypotheses. A hypothesis most consistent with the MDM is that comorbid dyslexia and DCD are the result of shared genetic risk factors which lead to the development of shared cognitive

impairments that contribute to dyslexia and DCD ('shared aetiology' hypothesis; de Jong, Oosterlaan, & Sergeant, 2006). The shared aetiology hypothesis supports the MDM as both suggest shared genetic factors are responsible for comorbid disorders. There is also converging evidence in the literature examining dyslexia and comorbid disorders that support a shared aetiology account (e.g., Gooch, Snowling, & Hulme, 2011; Moll et al., 2016 Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005; see also Cruddace & Riddell, 2006).

Alternative competing hypotheses of comorbid developmental disorders have also been postulated but have largely been discounted in comparisons between dyslexia and developmental disorders including ADHD (e.g., Gooch et al., 2011; see also Pennington, 2006) but not dyslexia and DCD. Competing hypotheses include the so-called 'phenocopy' hypothesis (Pennington, Groisser, & Welsh, 1993) whereby the behavioural symptoms of literacy difficulties (e.g., poor reading/spelling) are casually related to the child's motor difficulties or vice-versa. An alternative hypothesis, the 'cognitive subtypes' hypothesis, suggests comorbid dyslexia and DCD could be explained by the presence an entirely separable disorder to singular dyslexia and DCD (de Jong et al., 2006). Finally, comorbid dyslexia and DCD could be the result of an unknown third impairment (e.g., attention) which was not measured in this investigation (Scarborough & Dobrich, 1990).

4.4.4. Considerations in Using Handwriting Measures to Index Motor Skills

Methods for measuring motor skills in a large group of children simultaneously are limited. In deciding how to measure motor skills, we followed Lingam et al. (2009) by incorporating handwriting as a measure of motor skills used in daily activities. The use of handwriting difficulties as a criterion for DCD was also supported by the DSM-V (APA, 2013) where handwriting difficulties are identified as symptom of impaired daily motor activity in children with DCD.

However, handwriting is not just a motor skill and there is some evidence to suggest children with dyslexia also have handwriting legibility difficulties (Abbott & Berninger, 1993; Martlew, 1992). Therefore, it is conceivable, that some children with only literacy difficulties may have been misclassified as having comorbid literacy and motor difficulties due to their poor handwriting legibility. However, the incidence of this occurring was minimal because 81% of those identified in this study as having comorbid literacy and motor difficulties were later classified as having comorbid dyslexia and DCD using a larger battery of measures which did not include handwriting (see Study 2). Although the use of handwriting ability as an index of motor ability may have led to a small minority of children being incorrectly classified as having comorbid literacy and motor difficulties, the current battery's sensitivity for correctly identifying children with comorbid literacy and motor difficulties was above the 80% threshold for appropriate sensitivity of a developmental screening battery (Glascoe & Byrne, 1993).

It is also important to note that handwriting is a taught skill. The time spent and the quality of the instruction children receives varies somewhat between schools (Graham et al., 2008). Even though handwriting is included in the national curriculum (Learning Wales, 2014) it is likely that small between school effects identified in this study reflect inter-school differences on the duration and quality of handwriting (see Chapter 1).

4.4.5. The Utility of Literacy and Motor Skill Classroom Screening

This study used a novel approach to assessing literacy and motor skills concurrently in classrooms. Comparisons between the frequency of children with literacy and motor difficulties in this study with other epidemiological surveys (Lingham et al., 2009; Snowling and Hulme, 2015) employing different paradigms shows convergent findings in the base rates of literacy and movement difficulties. This convergence tentatively suggests that concurrent classroom screening of literacy and motor skills is valid. Further work is required to fully

examine the sensitivity and specificity of this screening battery though, particularly in reference to the motor tests where a focused set of motor skill related tasks were used.

4.4.6. Limiting Considerations

The primary limitation of this study is in the way motor skills were assessed. Unfortunately, at present, no suitable method for assessing motor skills in classes exist. Instead, we elected to use a test of visual motor integration and handwriting ability. The test of visual motor integration is a test of complex visuospatial skill that taps fine motor skills. Although perceptuomotor skills discriminate between children with DCD and typically developing children at the group level, some children with functional motor impairments who meet the criteria for DCD do not have perceptuomotor deficits (e.g., Wilson & McKenzie, 1998; Wilson et al., 2013). This means that this measure may not have been sensitive in identifying some children with functional motor impairments. Furthermore, our measures of motor skills predominantly assessed fine motor skills and not gross motor skills/balance meaning the current battery may not have been sensitive in detecting children who meet the diagnostic criteria for DCD because of gross motor/balance impairments. However, the similarities in frequency of DCD between the current study and previous epidemiological studies assessing a wider range of motor skills (e.g., Lingham et al., 2009) suggests that the battery was appropriate.

As noted earlier, we did not assess the prevalence of attentional impairments. Attentional disorders are frequently comorbid with both dyslexia (Willcutt et al., 2005) and DCD (Martin, Piek, & Hay, 2006). Furthermore, attentional impairments are causally linked to handwriting difficulties (Racine, Majnemer, Shevell, & Snider, 2008), motor disorders (Piek, Pitcher, & Hay, 1999), and explain DCD type deficits found in children with dyslexia (Wimmer et al., 1999). As we did not assess attentional skills in the current sample, it was not possible to address whether comorbid literacy and motor difficulties were related to attention

disorders. Moreover, it is possible that our prevalence estimates of singular and comorbid literacy and motor difficulties were inflated by contamination from children with attentional disorders. However, inflated estimates seem unlikely in this study as the current prevalence estimates of singular disorders match previous investigations and our estimates of comorbid difficulties were more conservative than other reported estimates.

4.4.7. Conclusion

Before examining transcription impairments amongst children with dyslexia and DCD, it was first necessary to investigate evidence of relationship between the disorders as is predicted in current models of the neurodevelopmental disorders (e.g., Pennington, 2006). To do so, we examined the prevalence of singular and comorbid literacy and motor difficulties in a large community sample. We found similar numbers of children with singular literacy and motor difficulties to other UK based studies and a higher frequency of children with comorbid literacy and motor difficulties than expected. Thus, the present findings, not only validate the use of a class screening paradigm to identify children with literacy and/or motor difficulties; but indicates that there is frequent comorbidity between the two disorders. This comorbidity further suggests a relationship between the two disorders. Yet, the nature of this relationship remains unclear.

Chapter 5

Study 2: Profiles of Phonological, Literacy, Perceptuomotor, and Executive Skills in Dyslexia, DCD, and Comorbid Dyslexia and DCD

5.1. Introduction

The literature reports a high degree of overlap of phonological, visuospatial, executive function, motor, and literacy deficits between dyslexia and DCD. A potential explanation for this overlap is the high frequency of comorbidity between the two disorders (Study 1). Yet, little is known about the nature of this comorbidity. In this study, we examine the profiles of children with dyslexia, DCD, and comorbid dyslexia and DCD to (a) disentangle independent and shared impairments of the disorders and (b) investigate the basis of comorbidity between the two disorders.

5.1.1. Considering Dyslexia and DCD in the Multiple Deficit Model

In Study 1 the frequency of comorbid literacy and motor difficulties was greater than chance which tentatively suggests a relationship between literacy and motor disorders. This provides some support for the prediction made by the multiple deficit model (MDM; Pennington, 2006) that neurodevelopmental disorders such as dyslexia and DCD share aetiological risk factors (see Introduction for full description of this model). In this study, the multifactorial nature of dyslexia and DCD is examined more closely.

The MDM hypothesises disorders such as dyslexia and DCD result from numerous aetiologically and cognitive risk factors that act in a probabilistic manner to increase the likelihood of an individual meeting a diagnostic threshold. Some of these risk factors are specific to a disorder, that is, they are independent, whilst others are shared between disorders. The presence of shared risk factors increases the likelihood of comorbidity between the disorders (Pennington, 2006). This hypothesis has led to a proliferation of studies investigating independent and shared risk factors of dyslexia (e.g., Gooch, Snowling, & Hulme, 2011; Moll, Göbel, Gooch, Landerl, & Snowling, 2016). To date, however, it remains unclear what the independent and shared risk factors of dyslexia and DCD are.

Studies investigating dyslexia and DCD separately have reported an overlap in deficits. The MDM distinguished between different levels of analysis: the aetiological, neural, cognitive, and symptom level. In the MDM model, the term symptom is largely equivalent to Frith's (1999) behavioural level and describes the impairments which define a disorder. In this context, the symptoms of dyslexia are literacy (reading and spelling) impairments (e.g., Rose, 2009) whereas the symptoms of DCD are motor impairments (e.g., APA, 2013). This study is concerned with the reported co-incidence of deficits in phonological, visuospatial, and executive abilities at the cognitive level and of the literacy and motor symptoms of the disorders between dyslexia and DCD (see Figure 5.1). In what follows, the literature reporting the apparent overlap in cognitive deficits and literacy and motor symptoms between dyslexia and DCD is critically evaluated.
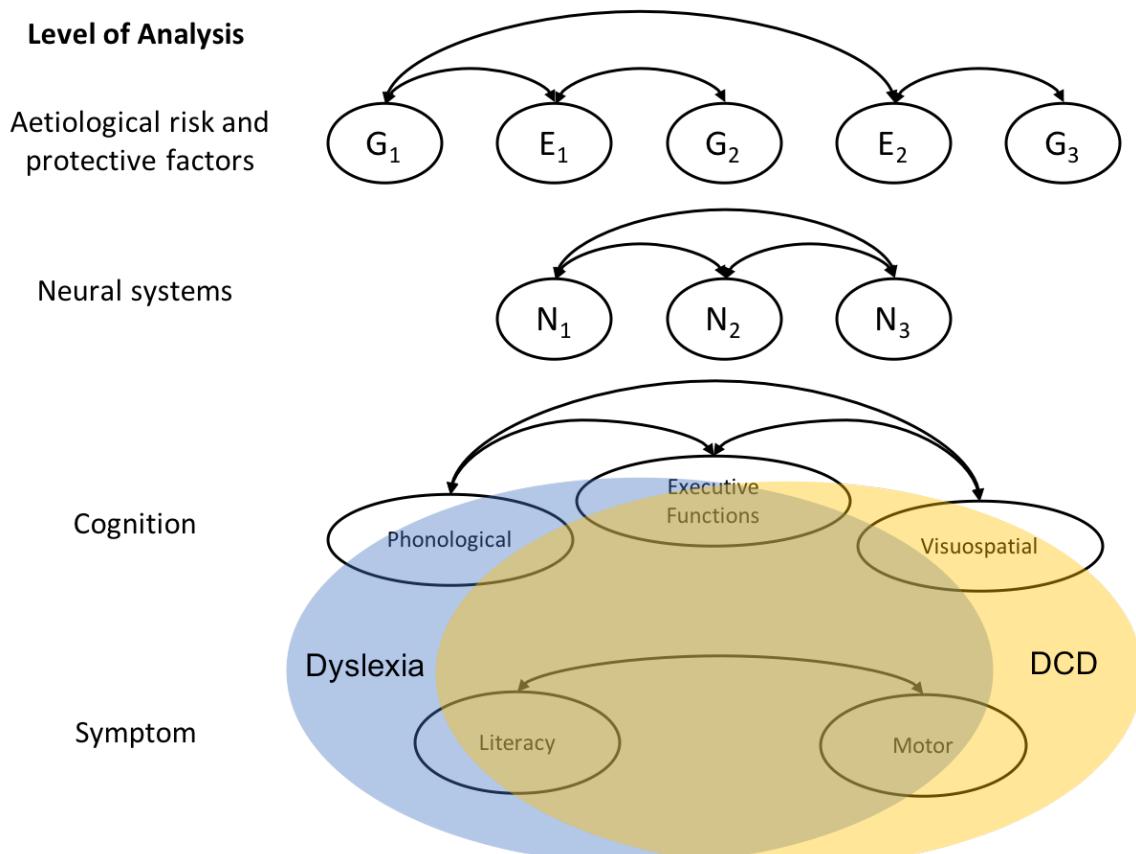
Figure 5.1. An adapted version of the multiple deficit model (Pennington, 2006) of the overlap in cognitive deficits and literacy and motor symptoms between dyslexia (blue) and DCD (yellow).

5.1.2. Overlap of Cognitive Deficits

5.1.2.1. Phonological deficits. As discussed earlier (see Chapters 1 and 2), it is well established that phonological skills are a critical determinant in learning to read and spell (Caravolas et al., 2012; Melby-Lervåg et al., 2012). Children with dyslexia typically experience phonological processing deficits (e.g., Snowling, 2000, Vellutino, Fletcher, Snowling, & Scanlon, 2004), which precede and predict their later literacy (dis)abilities (Dandache et al., 2014; Hulme, Nash, Gooch, Lervåg, & Snowling, 2015; Landerl et al., 2013; Moll et al., 2016; Pennington & Lefly, 2001). Interventions with children at risk of or experiencing dyslexia further demonstrate that training phonological skills improves their phonological *and* literacy ability (e.g., Hulme, Bowyer-Crane, Carroll, Duff, & Snowling, 2012). Thus, phonological deficits are common in dyslexia and are causally related to the disorder.

Notably, however, some children who have phonological deficits go on to develop typical reading and spelling skills, while others with poor literacy do not appear to have phonological deficits (Ramus et al., 2003; Snowling, 2008). Thus, phonological deficits by themselves may not be sufficient to cause dyslexia. Rather, phonological deficits act probabilistically with other cognitive deficits to increase the risk for a child to meet diagnostic criteria for dyslexia (Moll et al., 2016; Pennington, 2006). In this view, phonological deficits are a cognitive risk factor or marker which increases the risk of an individual being dyslexic.

Difficulties on measures which require phonological skills have also been reported amongst children with DCD. Children with DCD and suspected DCD were significantly less accurate than typically developing children when reading nonwords, a measure that taps phonological skills (Dewey et al., 2002). On a similar task (nonword repetition), children with DCD reproduced significantly fewer words correctly than typically developing controls.

In the DCD group, 45% of children scored at least 1 *SD* below their age average on the test (Archibald & Alloway, 2008). However, it is important to note that performance on nonword repetition tasks is also explained by oral language ability (e.g., listening comprehension and vocabulary) and so weak oral language skills might also explain low performance amongst children with DCD on nonword repetition tasks (Melby-lervåg & Lervåg, 2012). Furthermore, although nearly half the sample of children with DCD had poor performance on a phonologically related task, the proportion of children with DCD who had difficulties was smaller than that reported in dyslexic samples. For example, 77% of children with dyslexia in Ramus et al. (2003) scored 1 *SD* below their age average on phonological-related measures including nonword reading.

The variability of children with DCD on tasks that tap phonological skills suggests phonological deficits may represent some, but not all of children with DCD. The lack of studies (a) using more direct measures of phonological skills (e.g., phoneme deletion tasks) and (b) employing designs beyond case-controls (e.g., longitudinal) precludes a closer examination of the relationship between language and literacy development in children with DCD. A potential explanation for the presence of phonological deficits in DCD could be the inclusion of children with comorbid dyslexia and DCD in samples of DCD. Notably, Archibald and Alloway (2008) controlled for comorbid SLI but did not control for comorbid dyslexia. As such, they did not include measures of reading and spelling which would have enabled the identification of children with comorbid dyslexia. Dewey et al. (2002) did identify children DCD who also had poor reading and spelling in their sample but did not discriminate between children with phonological deficits who had literacy impairments and those who did not. Discriminating between children with DCD who have phonological deficits and literacy difficulties from those with phonological deficits without literacy

difficulties is important for understanding whether phonological deficits are specific to DCD

or simply a reflection of the high incidence of comorbidity between dyslexia and DCD.

    5.1.2.2. Visuospatial deficits in dyslexia and DCD. Perceptual skills are functional

in localising information and providing feedback for correction of goal directed movements

(e.g., Wolpert & Ghahramani, 2000), hence they are important for acquiring and making

skilled motor actions (Halsband & Lange, 2006; Jeannerod, 2006). They are so tightly

coupled, they are often referred to jointly as *perceptuomotor skills*. A perceptual skill

important for motor action is visuospatial processing which is important for localisation and

directed action (Jeannerod, 2006; Valyear et al., 2006).

    Children with DCD are impaired on visuospatial tasks regardless of whether they

require a motoric response or not. For example, children with DCD perform less well on

tasks without a motor component such as length judgement, visual discrimination, form

constancy, and picture closure (Hulme et al., 1982; Tsai et al., 2008; Wilson et al., 2013) and

on tasks with a motor component such as visual motor integration (Bonifacci, 2004;

Schoemaker et al., 2001; Van Waelvelde, De Weerdt, De Cock, & Smits-engelsman, 2004).

Indeed, a meta-analysis of these studies by Wilson et al. (2013) have shown that children

with DCD do substantially less well than children without DCD on complex visuospatial

tasks with a motor component ($d = 1.27$) and on visuoperceptual tasks without a motor

component ($d = 0.83$).

    Despite average large group effects on these tasks, the relationship between

visuospatial processing and DCD is unclear. Whilst some have found significant correlations

between visuospatial processing and functional motor skills in children with DCD (Lord &

Hulme, 1987; Tsai & Wu, 2008) others have reported no associations (Henderson et al.,

1994; Prunty et al., 2016). The mixed findings and lack of longitudinal and training

investigations examining the relationships between these abilities preclude strong claims

about the causal role of visuospatial processing deficits in DCD. Nevertheless, the strong

association between visuospatial skills and typical motor development and action as well as

the clear difficulties exerted by tasks involving visuospatial processing on children with DCD

suggest that poor performance on visuospatial tasks is likely to be a cognitive risk factor of

DCD.

Visuospatial deficits have also been reported amongst children with dyslexia (e.g.,

Stein, 2001). Recently, Bellocchi, Muneaux, and Huau (2017) assessed visuospatial ability in

children with dyslexia in two studies. In the first, they compared dyslexics to chronological-

age and reading-ability matched controls on measures of visuospatial processing (visual

perception and visual motor integration). At the group level, children with dyslexia had

significantly lower visual perception and visual motor integration scores than both control

groups. However, most of the dyslexics scored within 1 *SD* on the measures (55% and 75%

of dyslexic group on visual perception and visual-motor integration, respectively). Notably,

the authors failed to control for comorbid DCD in their sample suggesting that the low group

mean in the dyslexia sample could be attributable to a small number of children with

comorbid dyslexia and DCD.

This point is somewhat validated when looking at the second study of dyslexics

reported by Bellocchi et al. (2017). In this study, the authors compared performance between

children with dyslexia-only, DCD-only, and comorbid dyslexia and DCD on the same tests as

were used in their previous study, thus measuring and controlling for confounds of

comorbidity in the singular disorder groups. Children with dyslexia achieved significantly

higher scores than children with singular and comorbid DCD, who did not differ from one

another. Crucially, no child in the dyslexia-only group scored less than 1 *SD* below their age

average, suggesting that deficits in visuospatial abilities observed in those with dyslexia in

Bellocchi et al.'s (2017) previous study represented cases of comorbid DCD. The lack of a

typical control group in the second study makes it difficult to rule out the presence of a sub-clinical visuospatial processing deficit in dyslexia completely, however.

5.1.2.3. Executive Function deficits. Executive skills are domain general cognitive processes necessary for supervising behaviour (Diamond, 2013). There are several cognitive processes which fall under the umbrella of executive functions (Cirino et al., 2018) and this study focuses on two commonly measured executive functions, memory and selective attention.

5.1.2.3.1. Memory. Memory itself is not unitary process and it is important to note that there are conflicting views whether short-term memory and working memory are separate systems or not. Some theorists assert short-term memory and working memory are separable constructs whilst others argue short-term memory reflects the 'slave' component of working memory (see Swanson, Zheng, & Jerman, 2009). Investigation of the structures of memory is beyond the scope of this thesis, and as such, memory here refers to both short-term and working memory unless stated otherwise.

The investigation of memory deficits in dyslexia has been the subject to a large volume of work (see Kudo, Lussier, & Swanson, 2015; Swanson et al., 2009 for meta-analyses). Deficits in verbal working memory are often reported amongst children with dyslexia (Kudo et al., 2015; Ramus, Marshall, Rosen, & van der Lely, 2013; Ransby & Lee Swanson, 2003), albeit verbal memory deficits are not as large as phonological deficits (Melby-Lervåg et al., 2012). Some investigations also report visual memory deficits (e.g., Varvara, Varuzza, Sorrentino, Anna, Vicari, & Menghini, 2014), however, others find no evidence of the latter (e.g., Moll et al., 2016). In their meta-analysis, Swanson et al. (2009) found visual memory impairments were no longer significantly related to dyslexia when accounting for math ability (e.g., van der Sluis, van der Leij, & de Jong, 2005). Thus, after

controlling for comorbid difficulties such as math disorder, verbal rather than visual memory deficits are most associated with dyslexia.

Whilst memory deficits in dyslexia seem to be restricted to the verbal domain, memory deficits amongst children with DCD appear to be more diffuse with a greater severity in the visuospatial domain (Blank et al., 2012; Wilson et al., 2013). Much of the evidence identifying memory deficits amongst children with DCD comes from Alloway and colleagues (Alloway, 2011; Alloway & Temple, 2007; Alloway, 2007; Archibald & Alloway, 2008). Their studies appear to involve the same sample of 55 children with DCD who were tested using the Automated Working Memory Assessment (AWMA; Alloway, Gathercole, & Pickering, 2004). Alloway (2007) analysed memory performance in a DCD-only group and reported low composite scores across the verbal and visual domains. Furthermore, visual memory scores were significantly lower than verbal short-term memory, suggesting larger deficits in visual memory in DCD. It is important to note that only 60% of the sample scored < 1.5 *SD* their age average on tests of visuospatial memory, suggesting visuospatial memory deficits are common, but are not characteristic of the disorder.

In addition to the work by Alloway and colleagues, other research groups have identified memory deficits in children with DCD (Crawford & Dewey, 2008; Parush et al., 1998; Tsai et al., 2008). These authors identified memory deficits using the Visual Memory and Sequencing Memory subtests from the Test of Visual Perceptual Skills (TVPS; Gardner, 1988). Both these subtests and the visual memory subtests used in the AWMA tap visuospatial processing as well as memory. For instance, in the Visual Memory Test children must recall a form or sequence from several distractor items after a five second delay. Selecting the correct target from several distractors requires visuospatial processing in addition to the memory dimension added by the delayed recall component. Given that visuospatial deficits are present in DCD (Wilson et al., 2013), it is possible that memory

deficits observed on these tasks were confounded by the visuospatial processing component of the task. In establishing a basis for a memory deficit amongst children with DCD it is important to consider what additional processes memory tasks tap and to control for potential extraneous effects through task selection or statistical procedures.

From the evidence reviewed here, it is apparent that memory deficits are present in both dyslexia and DCD. In dyslexia, memory deficits appear to be restricted to the verbal domain whilst in children with DCD they also appear to be more generalised. Notwithstanding issues on the testing for memory impairments, deficits in memory could be indicative of shared executive function deficits in dyslexia and DCD.

5.1.2.3.2 Attention. Like memory, attention is not a unitary process. According to one view, attention is divided into three sub-processes, sustained, selective, and control (Manly et al., 2001; Shapiro, Morris, Morris, & Jones, 1998). Deficits in attentional processes are most commonly are associated with ADHD (Barkley, 1997; Castellanos & Tannock, 2002; Manly et al., 2001) which is also frequently comorbid with dyslexia and DCD (Germanò et al., 2010; Kadesjö & Gillberg, 1999).

Selective attention, however, is unrelated to ADHD (Manly et al., 2001) but impairments on tasks of selective attention have been reported amongst children with dyslexia (Casco, Tressoldi, & Dellantonio, 1998; Menghini, Finzi, Benassi, Bolzani, & Facoetti, 2010; Varvara et al., 2014) and DCD (Wilson, Maruff, & McKenzie, 1997; Wilson & Maruff, 1999). An investigation directly comparing profiles of selective attention amongst children with dyslexia, DCD, and comorbid dyslexia and DCD found all disorder groups achieved roughly equivalent low scaled scores (~ 7). The control group also received a relatively low and statistically similar scaled score (8; Cruddace & Riddell, 2006). Therefore, it is possible that the measure was not sensitive enough to detect selective attention deficits,

or, selective attention deficits do not characterise these groups and should be investigated further.

Memory and attention have been discussed individually here. Whilst they represent separate processes, they also provide an indirect measure of executive functions. There is some evidence to suggest that executive function deficits may be present in dyslexia and DCD. In establishing whether deficits in executive functions are present in dyslexia and/or DCD, it is necessary to rule out potential confounds such as uncontrolled comorbidity and measurement issues (e.g., measures with poor sensitivity). Executive function deficits – in the absence of potential confounds – are unlikely to be direct causes of literacy or motor impairments associated with dyslexia and/or DCD but could interact with disorder-specific deficits to compound impairments at the behavioural level and increase the likelihood of a child receiving a diagnosis (Hulme & Snowling, 2009; Gathercole et al., 2016; Miller et al., 2014).

5.1.3. Co-incidence of Literacy and Motor Symptoms

In addition to the apparent overlap in cognitive deficits, studies examining dyslexia and DCD separately have reported an overlap in behavioural symptoms between the disorders. That is, children with dyslexia have been found to have motor impairments and children with DCD have been found to have literacy impairments.

5.1.3.4. Motor impairments and dyslexia. Impairments in motor functions as a consequence of cerebellar dysfunction, namely balance, have been claimed to characterize children with dyslexia by proponents of the *automatisation deficit hypothesis of dyslexia* (Nicolson & Fawcett, 1990; Nicolson, Fawcett, & Dean, 2001; see Chapter 2, pp. 46 - 50). The central tenet of this theory is that children with dyslexia have difficulties in maintaining balance whilst completing a secondary task. Subsequent studies have failed to replicate these findings and established that balance impairments more likely reflect comorbid disorders

such as ADHD (Raberger & Wimmer, 2003; Ramus et al., 2003; Rochelle & Talcott, 2006).

For example, children with ADHD-only and comorbid ADHD and dyslexia were impaired on

a balancing task similar to the one used by Nicolson and Fawcett (1990), yet children with

dyslexia-only were not (Raberger & Wimmer, 2003). Thus, balance difficulties were found to

be an impairment of ADHD and not dyslexia. This conclusion was also reported in a meta-

analysis of studies reporting balance difficulties amongst children with dyslexia. The meta-

analysis found balance impairments reported in children with dyslexia were most likely the

result of a third variable influence, such as comorbid ADHD (Rochelle & Talcott, 2006).

Researchers investigating the automatization deficit hypothesis have predominantly

focused on assessing motor skills predicted to be impaired by cerebellar dysfunction (balance

and fine motor skill) and have not tested performance on other aspects of motor skills (e.g.,

gross motor skills). A study by Iversen, Berg, Ellertsen, and Tønnessen (2005) examined

global aspects of motor skill. In this study children with dyslexia were examined on a broad

range of motor impairments (fine motor skill, gross motor skill, and balance). Interestingly,

children with dyslexia achieved significantly lower scores than controls on tests of fine motor

skills and balance but not on gross motor skills. Over half of the dyslexic group scored below

the 5th centile on the global score on the M-ABC, partially fulfilling diagnostic criteria for

DCD (see Chapter 2, p. 50) which suggests the motor difficulties at the group level might

best be explained by the presence of children with comorbid DCD in the sample. If fine

motor and balance impairments in this sample were due to comorbid cases, then the data

suggest a profile of specific motor impairments in comorbid dyslexia and DCD. However, the

exact nature of motor impairments in comorbid dyslexia and DCD remains unknown.

5.1.1.5. Literacy impairments and DCD. It has also been reported that children with

DCD have lower reading and spelling skills, yet ability is highly variable. For example, in the

ALSPAC population study described in Study 1, only children with severe DCD (< 5th centile

on a motor assessment battery) had significantly lower reading and spelling ability

(Schoemaker et al., 2013), whilst in another study with a smaller sample (N = 430) children

with mild DCD (< 25th centile on a motor assessment battery) had significantly lower reading

and spelling ability than typically developing children (Dewey et al., 2002). In the latter

study, there was a larger amount of variability in literacy ability among the DCD group than

in the controls. This within-DCD variability was also apparent in Alloway (2007) where 46%

of children with DCD were more than 1 *SD* below their age average on a composite measure

of literacy (including word reading and spelling). Such variability suggests literacy

difficulties exist in some but not all of the group. Again, one possibility is that the variability

reflects the inclusion of children with comorbid dyslexia and DCD in the samples.

Studies investigating phonological, visuospatial, executive functions, literacy, and

motor impairments in dyslexia and DCD have predominantly examined either dyslexia or

DCD but not both. The findings from these studies suggest a high degree of overlap in

cognitive deficits as well as behavioural symptoms. However, it is unclear in the vast

majority of cases whether researchers have controlled for high frequency of comorbidity

between the disorders (see Study 1). It is therefore timely to examine whether the overlap in

impairments between dyslexia and DCD remain after identifying and controlling for

comorbidity. Doing so, will delineate the profiles of cognitive deficits and symptoms

between dyslexia and DCD and allow the identification of independent and shared deficits

between the disorders.

Following Pennington's (2006) terminology, deficits in cognitive abilities relating to

either one or both disorders should be regarded as a cognitive risk factor. However, because

we are investigating overlap in both cognitive deficits and symptoms (see Figure 5.1) and are

therefore not investigating deficits strictly in the cognitive domain, the term *marker* will be

used here to describe both cognitive deficits and literacy and motor impairments. As such, the

first aim of this study is to identify independent and shared markers of dyslexia and DCD, which is critical for understanding the aetiology of the disorders, including their frequent comorbidity.

5.1.4. Comorbidity between Dyslexia and DCD

Despite the high frequency of comorbidity between dyslexia and DCD (Study 1) and the implications for uncontrolled comorbidity discussed in the preceding section, the nature of this comorbidity remains unclear. Comorbidity between dyslexia and DCD crosses separate language and motor domains and is therefore regarded as a heterotypic comorbidity (Angold et al., 1999). An example of a well-researched heterotypic comorbidity is that between dyslexia and ADHD. Double dissociation studies of singular and comorbid dyslexia and ADHD have produced several accounts for the genetic aetiology of comorbidity (de Jong et al., 2006; Pennington, 2006). Such accounts may also apply to comorbidity between dyslexia and DCD by providing competing predictions of the profiles of impairments between the singular and comorbid groups. The three accounts which have received the most attention, phenocopy, cognitive subtypes, and shared aetiology hypotheses, will be discussed in turn, with a focus on what each account would predict when examining the comorbidity between dyslexia and DCD.

5.1.4.1. Phenocopy. The phenocopy account suggests that a single aetiology causes cognitive deficits consistent with a singular disorder but these deficits lead to behavioural manifestations of a second disorder (Pennington et al., 1993). A so-called *copy* of the symptoms at the behavioural level. Evidence for this account rests most strongly on a double dissociation study by Pennington et al. (1993). In this study, the authors found children with dyslexia-only had a phonological processing deficit but spared executive functions whereas children with ADHD-only had an executive function deficit with spared phonological processing. In a similar manner to children with dyslexia-only, children with comorbid

dyslexia and ADHD had phonological processing deficits but spared executive functions. Pennington et al. (1993) reasoned that the behavioural impairments related to ADHD in the comorbid group were a downstream product of reading difficulties. However, later studies have failed to replicate the phenocopy hypothesis in dyslexia and ADHD. These studies found the comorbid group profile reflects a combination of deficits associated with both dyslexia and ADHD (Willcutt et al., 2001). If the phenocopy hypothesis were true in the case of comorbid dyslexia and DCD, deficits associated with either dyslexia or DCD but not both disorders would be present in the comorbid group.

5.1.4.2. Cognitive subtype. The cognitive subtype hypothesis suggests comorbidity stems from a separate aetiology to either of the singular disorders (de Jong et al., 2006). According to this view, two separate aetiologies are responsible for each singular disorder and a third aetiology is separately responsible for the comorbid disorder. Evidence for the cognitive subtype hypothesis comes from studies that find children with comorbid disorders had different or more severe deficits as compared to either singular disorder group (Mcgee, Brodeur, Symons, Andrade, & Fahie, 2004; Rucklidge & Tannock, 2002; Willcutt et al., 2001).

When comparing children with dyslexia and/or ADHD on measures of inhibition, set-shifting, working memory, and phoneme awareness, Willcutt et al. (2001) found that children with singular dyslexia were impaired on tasks of working memory and phoneme awareness whereas children with singular ADHD had deficits in inhibition. The comorbid group were more impaired than either singular disorder on measures of inhibition and phoneme awareness. The dissociation in profiles between the comorbid and either singular disorder group indicates a separate, or third, genetic basis. If the cognitive subtype hypothesis were applicable in the case of comorbid dyslexia and DCD, children with comorbid dyslexia and

DCD would have a different profile and/or more severe deficits than either dyslexia-only or DCD-only groups combined.

      5.1.4.3. Shared aetiology. According to the third and final hypothesis, both disorders have a partially common genetic basis capable of producing either or both singular disorders (de Jong et al., 2006). In this case, the comorbid disorder would represent an additive combination of deficits associated with the singular disorders with the severity of deficits being no different in the comorbid disorder than a combination of both the singular disorders. Recent work has found evidence for this hypothesis in other heterotypic comorbidities including dyslexia and ADHD (Gooch et al., 2011; Willcutt et al., 2005) and dyslexia and math disorder (Moll et al., 2016). Gooch et al. (2011) examined performance between groups of singular and comorbid dyslexia and ADHD on their phonological skills, executive function, and time perception (a deficit associated with both dyslexia and ADHD). Children with singular dyslexia had deficits in phonological skills and time perception whereas children with singular ADHD had deficits in executive function and time perception. The comorbid group were deficient on all three measures, but these deficits were no more severe than either singular disorder, consistent with a shared aetiology account. Accordingly, this account would predict children with comorbid dyslexia and DCD would have a similar profile of deficits – not differing in severity – to singular dyslexia and DCD.

      Studies examining the comorbidity between dyslexia and DCD have reported findings consistent with the cognitive subtype and shared aetiology hypotheses. Crawford and Dewey (2008) compared children with singular and comorbid dyslexia, DCD, and ADHD on tests of visuospatial processing (TVPS; Gardner, 1988) and Rey-Osterreith Complex Figure (Ostereith, 1944). On the visual memory subtest of the TVPS and on the Rey-Osterreith Complex Figure, children with comorbid dyslexia, DCD, and ADHD performed significantly less well than the singular disorder groups, implying a cognitive subtype aetiology. However,

on other measures, such as the TVPS total score, children with comorbid dyslexia, DCD, and ADHD did not differ significantly from the singular dyslexia group, implying a shared aetiology. On the other hand, Biotteau et al. (2015) found no differences between singular and comorbid dyslexia and DCD on a procedural learning task, also supporting a shared aetiology hypothesis (see also Bellocchi et al., 2017). Thus, there appear to be inconsistencies in the manifestations of comorbidity between dyslexia and DCD, with no study using a double dissociation paradigm to assess the comorbidity between the disorders. As such, the second aim of this study was to examine the nature of comorbidity by testing the three – phenocopy, cognitive subtype, and shared aetiology – hypotheses of comorbidity.

5.1.5. The Current Study

The literature reports an overlap in the cognitive deficits and behavioural symptoms of dyslexia and DCD. A potential explanation for this overlap is the presence of children with comorbid dyslexia and DCD in samples. Indeed, the nature of comorbidity between dyslexia and DCD is also unclear. These issues were probed in this study by addressing two aims. The first was to examine profiles of performance on tests of phonological, visuospatial, executive functions, literacy, and motor abilities to identify the independent and shared markers of dyslexia and DCD. The second aim was to investigate the profiles of children with comorbid dyslexia and DCD relative to children with dyslexia-only and DCD-only to establish the nature of comorbidity between these disorders.

5.1.5.1. Independent and shared markers of dyslexia and DCD. Based on the evidence reviewed, it was expected that children with dyslexia would have poorer phonological and literacy abilities, but their visuospatial and motor abilities would not differ from controls. In contrast, children with DCD would have poorer visuospatial and motor abilities but their phonological and literacy abilities would not differ from controls. Lower executive functions abilities would be identified in both dyslexia and DCD.

5.1.5.2. Comorbidity in dyslexia and DCD. It was expected children with comorbid dyslexia and DCD's pattern and severity of deficits would be similar to children with dyslexia-only and/or DCD-only on all abilities. That is, children with comorbid dyslexia and DCD would have an additive profile of deficits consistent with the shared aetiology hypothesis.

5.2. Methods

5.2.1. Participants

Children identified as being 'at risk' of literacy, motor, co-morbid literacy and motor difficulties, along with a random sample of children who were not identified as being at risk of literacy and/or motor difficulties in Study 1 were invited to take part in this study. Of those invited, a total of 141 children and their parents agreed to continue participating. Children were now in Years 4 ($n = 41$, 53% female), 5 ($n = 52$, 41% female), and 6 ($n = 48$, 44% female). A further 17 children also in Years 4 ($n = 7$), 5 ($n = 3$), and 6 ($n = 7$) who attended other mainstream primary schools in North Wales and were referred by education and health services in the area also took part.

Regardless of their categorisation in the previous study, children were re-classified as having dyslexia, DCD, and comorbid dyslexia and DCD using marker approach similar to the one used in Study 1. The marker criteria were applied to tests of literacy and motor skills administered in part during Study 1 and in the current investigation. The classification for dyslexia, DCD, comorbid dyslexia and DCD, or typical developing groups is outlined below.

5.2.1.1. Classification of dyslexia. Dyslexia was determined when children scored $\leq$ 1.33 *SD* relative to their age average on at least two tests of the following measures. Wide Range Achievement Test-IV (WRAT-IV) Word Reading and Spelling (Wilkinson & Robertson, 2006), Sentence Spelling (Caravolas, Volín, & Hulme, 2005), ELDEL One Minute Word Reading (Caravolas et al., 2012).

5.2.1.2. Classification of DCD. DCD was identified when children scored ≤ 1.33 *SD* relative to their age average on at least two tests of the following four subtests from the Motor Assessment Battery for Children 2 (M-ABC 2; Henderson, Sugden, & Barnett, 2007): Trail Drawing, Threading, Bag Throw, and One Board Balance.

5.2.1.3. Classification of comorbid dyslexia and DCD. Children were classified as having comorbid dyslexia and DCD when they met the above criteria for both dyslexia and DCD.

5.2.1.4. Classification of typically developing (TD). Children who did not meet any of the criteria for the three previous 'disorder groups' were classified as being typically developing.

5.2.1.5. Sample characteristics. The characteristics of each of the four groups is reported in Table 5.1. Groups did not differ in age but children with comorbid dyslexia and DCD had significantly lower scores on the measure of general non-verbal ability (NVIQ), measured using the WISC–IV Block Design subtest (Wechsler, 2004). Children with dyslexia (singular and comorbid) had lower scores on all literacy measures, as expected. On the motor measures, children with DCD (singular and comorbid) had lower scores. Children with dyslexia also scored slightly lower on the measure of gross motor skills, Bag Throw. Thus, it appears children with DCD were not impaired on literacy skills, but children with dyslexia may have some small (sub-clinical) impairments in motor skills, without meeting the criteria for DCD.

Table 5.1.

Performance on Selection Tests by Children with Dyslexia, DCD, Dyslexia and DCD with Typically Developing Children Controls

	DYS		DCD		DYS + DCD		Typically Developing				Post-Hoc Comparisons[†]		
	M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*F*	η^2_ρ	TD vs Dys	TD vs DCD	TD vs Dys+DCD
n	28		25		17		83						
% Female	32		32		59		49						
Age (months)	118.04	10.29	118.24	10.47	119.35	12.97	118.33	11.03	0.06	.00			
Non-verbal IQ[a]	9.89	3.71	8.52	2.63	6.60	2.59	9.97	3.30	5.36**	.10			**
Sustained Attention[a]	8.96	2.95	7.76	3.90	7.24	2.36	8.86	3.55	1.64	.03			
Literacy Skills													
Sentence Spelling[b]	22.56	7.32	38.28	10.42	19.41	7.32	41.61	9.81	67.45***	.49	***		***
Word Spelling[c]	78.54	9.34	103.92	15.07	73.82	8.56	106.94	14.75	51.90***	.51	***		***
Word Reading[c]	82.46	8.40	100.80	12.88	82.06	7.15	101.81	9.86	39.98***	.45	***		***
One Minute Word Reading[b]	67.44	14.61	88.39	15.51	68.93	19.12	96.32	13.88	32.64***	.42	***		***
Motor Skills													
Shape Tracing[c]	90.29	6.78	75.20	9.76	78.06	10.79	92.12	8.82	30.74***	.38		***	***
Threading[a]	9.22	2.28	7.04	2.99	5.27	2.28	9.61	2.79	14.28***	.24		***	***
Bag Throw[a]	7.18	2.92	6.40	2.42	5.88	2.80	9.12	2.84	11.19***	.19	*	***	***
Balance[a]	9.93	2.45	9.12	3.53	7.76	2.54	10.74	2.39	7.15***	.13		*	***

Note. See Measures for description of the selection tests. DYS = dyslexia. DCD = Developmental coordination disorder. DYS+DCD = Comorbid dyslexia and developmental coordination disorder (Dyslexia + DCD). Non-verbal IQ was measured by the WISC-IV Block Design.
[a]Scaled scores. [b]Raw scores. All other scores were [c]standard scores.
[†]Post-hoc comparisons with Bonferroni corrections.

5.2.2. Design and Procedure

Children completed a large battery of multiple measures assessing phonological speed, literacy, visuospatial, motor, and executive function skills. Children who took part in the earlier study, completed testing sessions between October 2015 and December 2016 whilst children who were referred from specialist services were tested between September 2016 and May 2017 (see General Methods).

Testing was spread over seven sessions to reduce the likelihood of fatigue and boredom. Within each of the testing sessions, the administration order of the individual tests was fixed and manipulated to minimise the likelihood of transfer, or priming, from one test to the other (e.g., phonological speed tasks were interspersed within session one; see Table 5.2). Each testing session lasted no longer than one hour, and children were given an opportunity to take a short break after each test. Most of the tests were administered in Sessions 3 and 4. From Session 5 onwards, children completed additional tests and some experimental tasks not discussed in the current study. From Session 3 onwards, each child worked on a one-to-one basis in a quiet area of their school with a researcher (some children referred from specialist services were tested at the University lab). Published administration and scoring instructions, including any discontinue criteria, were followed.

Table 5.2.

Order of Test Administration and Primary Skill Domain the Measures were Testing

Session	Test	Skill Domain
2	Sentence Spelling (Caravolas et al., 2005)	Literacy
	Word Spelling (WRAT IV)	Literacy
	Visual Motor Integration (Beery VMI-VI)	Visuospatial
3	Phoneme Deletion (Caravolas et al., 2005)	Phonological Speed
	Visual Perception (Beery VMI-VI)	Visuospatial
	Verbal (Digit) Span (WISC-IV)	Verbal Memory
	One Minute Nonword Reading (Caravolas et al., 2005)	Phonological Speed
4	Word Reading (WRAT-IV)	Literacy
	RAN (Caravolas et al., 2012)	Phonological Speed
	Digits	
	Letters	
	Shape Tracing (Beery VMI-VI)	Motor
	Corsi Blocks (WMTB-C)	Visual Memory
	Sky Search (TEA-Ch)	Selective Attention
	Sky Search DT (TEA-Ch)	Selective Attention
5	Bag Throw (M-ABC 2)	Motor
	Balance (M-ABC 2)	Motor
6	Block Design (WISC-IV)	Nonverbal IQ
	Trail Drawing (WMTB-C)	Motor
7	Threading (WMTB-C)	Motor
	One Minute Word Reading (Caravolas, 2017)	Literacy

Note. See Measures for description of the selection tests. Tests given during Sessions 1 and 2 were administered to whole classes. In the rest of the sessions (3 – 7) tests were administered individually. The order only includes the tests relevant to this study and excludes the order of administration of tasks used in later studies. No tests relevant to this study were administered in Session 1. Experimental tasks were administered from Session 3 onward and were normally administered after the tests listed above.

5.2.3. Measures

Children completed a large battery of measures assessing the phonological, literacy, visuospatial, motor, and executive function skills. Tests were selected based on their use in previous studies to measure the same constructs of interest here and/or if they were theoretically linked to the respective constructs. Accordingly, descriptions of individual tests are given under their respective domains. In cases where the test was administered in Study 1 (e.g., Word Spelling) the test is briefly described again for reference.

5.2.3.1. General Ability. Children completed the Block Design subtest from the Wechsler Intelligence Scale for Children IV (WISC-IV; Wechsler, 2004) to measure non-verbal IQ (NVIQ). Arguably Block Design taps visuospatial motor skills, likely to be impaired in children with DCD. However, Block Design is reported not to discriminate between children with and without DCD (Sumner, Pratt, & Hill, 2016), as also corroborated by our analyses (Table 5.1), making it a suitable measure of nonverbal IQ here. During the task, the child used cubes with red, white, and half-red-half-white faces to reproduce designs displayed on paper in front of him/her within a time limit. Over 14 trials lasting between 75 and 120 seconds, the designs increased in complexity as well as the number of cubes required to complete them from four to nine. Correct trials were awarded four points with bonus points given on later trials if the child completed the designs in a shorter time. The task was discontinued if the child was unable to reproduce the design within the required time limit or made an error on three consecutive trials (maximum raw score = 68).

5.2.3.2. Phonological Speed. Speed of phonological processing was measured using Phoneme Deletion and Rapid Naming tests.

5.2.3.2.1. Phoneme Deletion. As in Caravolas et al. (2005), children were asked to remove the initial (Block 1), or final phoneme (Block 2) of aurally presented non-words. In Block 1 the child repeated the whole item before pronouncing the new one without the initial (onset) phoneme (e.g., 'roth' → 'oth'). The same procedure was followed in Block 2 where the child removed the final (coda) phoneme (e.g., 'koot' → 'koo'). Item complexity was manipulated such that half (n = 5) of the onset and coda items were of single consonants ('**r**oth', 'koo**t**') and half of two consonants ('treen', 'ke**st**'). For reliability, each half-block of five items was timed from the administrator's presentation of the first item until participant's production of the final response. The duration was the summed number of seconds taken to complete blocks one and two (across a total of 20 items).

5.2.3.2.2. Rapid Automatized Naming. Two variants of the RAN task – *digits and letters* – from Caravolas et al. (2012) were administered. In each case, the child was initially asked to name the five test stimuli to ensure familiarity. Afterwards, the child named the stimuli repeated pseudo-randomly in two, arrays of eight by five from left to right following with their finger as fast as they could. During the RAN Digits subtest children were asked to name the digits: 2, 3, 6, 7, and 9. In the RAN Letters subtest, children were asked to name the lowercase letters: a, d, p, o, and s. The time in seconds taken to name the items in each trial was recorded and then averaged over the two trials, as was the error (number of items named incorrectly or skipped, maximum error score = 40).

5.2.3.3. Literacy. In addition to the word and sentence spelling tests administered in Study One, reading accuracy and fluency was measured as part of the larger battery in this study.

5.2.3.3.1. Word Reading. The Wide Range Achievement Test IV Reading (WRAT-IV; Wilkinson & Robertson, 2006) was used to assess reading accuracy. During the test, each child was asked to read aloud from a 55-item graded word list. Words increased in difficulty and administration was discontinued after the child made ten consecutive errors. If the child made an error on any of the first five items, word reading was paused whilst the child read a basal set of 15 individual alphabet letters aloud. Accuracy was the number of correctly read letters and words prior to the discontinue rule (maximum raw score = 70).

5.2.3.3.2. Reading Fluency. Children completed two versions of a reading fluency test. In the *One Minute Word Reading* test (Caravolas et al., 2012), the child read aloud as many words as s/he could from a list of 144 high frequency words in 60 seconds. The words increased in length (one to eight letters) and in syllable number (one to three syllables).

In the *One Minute Non-Word Reading test* (Caravolas, 2017)*,* following the same procedure as above, the child read aloud from a list of 144 non-words as fast as they could in

1 minute. Non-words were derived from each of the word items administered in the One Minute Reading test by exchanging the first or last phoneme of syllables with a plausible alternative (e.g., 'cat' → 'jat'). It is important to note that particularly on the non-word reading test, children must rapidly process grapho-phonological information and so this test taps phonological processing skills to a greater extent than other reading and spelling measures administered. On both tests, errors and self-corrections were noted. Fluency on each test was the number of correctly read items in 60 seconds (maximum = 144).

5.2.3.3.3. Word Spelling. The Spelling subtest of the Wide Range Achievement Test IV (WRAT-IV; Wilkinson, & Robertson, 2006) administered in Study 1 was used. In this test, children wrote 13 alphabet letters and 36 words – graded in difficulty – to dictation. Scoring followed the published guidelines and was discontinued after 10 consecutive errors (maximum = 49).

5.2.3.3.4. Sentence Spelling. The sentence spelling scores (Caravolas et al., 2005) administered during Study 1 provided a secondary index of spelling ability. In this test, children wrote 10 sentences – graded in difficulty – to dictation. Scoring followed the guidelines and one point was awarded for every correct spelled word (maximum = 63).

5.2.3.4. Visuospatial ability. Perceptuomotor abilities underlie coordinated movements. In this study, we measured visual perception and visual motor integration. No test of kinaesthetic awareness was administered due to concerns about test's reliability and validity (Siringu & Gomez, 2015). The Beery VMI VI (Beery & Beery, 2010) administered as part of Study 1 was used to index visual motor integration. Visual perception was measured using another task from this battery.

5.2.3.4.1. Visual Motor Integration. Scores from Beery VMI (2010) administered during Study 1 was used as the index of visual motor integration in this study. Children

copied 24 forms of increasing complexity. Only one attempt per form was permitted and scoring followed the published guidelines (maximum = 30).

5.2.3.4.2. Visual Perception. A shape matching test from Beery and Beery (2010) was used to assess visual perception. The same 27 target shapes from the Visual Motor Integration test (Beery and Beery, 2010) were displayed above vertically organised distractors as well as the identical shape to the target. The complexity of the shapes increased as did the number of distractor shapes from two to seven. Children were asked to find and mark the shapes that matched their respective targets and were encouraged to be accurate rather than fast when doing so. Accuracy was the number of correctly matched shapes – prior to the discontinue rule of three consecutive errors – in 3 minutes (maximum raw score = 30).

5.2.3.5. Motor skills. To capture a range of motor skills, sub-tests from the both Beery VMI (Beery and Beery, 2010) and M-ABC 2 (Henderson, Sugden, & Barnett, 2007) were used. Fine motor coordination was measured using a Shape Tracing test by Beery and Beery (2010). In addition, we also used four subtests from age band two (7 – 11 years old) of the M-ABC 2 to measure fine motor, gross motor, and balance skills. Due to time constraints, we were limited in the measures that could be administered and so the subtests – Threading, Trail Drawing, Bag Throw, and One Board Balance – were selected because of theoretical motivations of how likely the individual tests were to capture specific motor skills (e.g., gross motor skill) and the reported sub-tests' factor loading on their respective constructs (see Schulz, Henderson, Sugden, & Barnett, 2011).

5.2.3.5.1. Shape Tracing. A third test by Beery and Beery (2010) was used to assess fine motor coordination. Each child was instructed to trace the 27 shapes – used in the previous tests by Beery and Beery (2010) – by keeping their pencil within the border lines of each shape. The first 19 shapes in the sequence included dots positioned at key junctions to make the shape easier to trace. As shapes increased in complexity, the distance between the

border lines decreased. Each response was scored on three criteria as described in the manual which included (a) whether there were pencil marks in all areas of the shape, (b) the pencil trace does not cross the border lines, and (c) the pencil trace must not cross overlapping joins on two of the latter complex shapes. Accuracy was the number of shapes drawn within 5 minutes that met the criteria described in the manual (maximum raw score = 30).

5.2.3.5.2. Trail Drawing. A second test of fine motor coordination was administered from the M-ABC 2. Children were asked to draw a continuous trace inside two boundary lines or 'trail'. Drawing began at a picture of bicycles and followed a zig zag path before it ended at a picture of a house. Towards the end of the trail, the distance between the lines narrowed. After a short demonstration and practise, the child completed two attempts of the trail without feedback from the administrator. Scoring of errors followed the comprehensive instructions outlined in the manual. The raw score was the number of errors from the trial with the fewest errors.

5.2.3.5.3. Threading. Fine motor fluency was assessed using the Lace Threading subtest from the M-ABC 2 (Henderson, Sugden, & Barnett, 2007). On the instruction of the administrator the child had to thread a string back and forth through eight holes drilled into a small plastic board. The task was timed from when the child's hands – positioned on the table either side of the board – left the mat, until they had pulled the string tight through the final hole. The threading time was the fastest time of two consecutive attempts.

5.2.3.5.4. Bag Throw. From a distance of 1.8 meters, the child threw a beanbag into a circular target (diameter = 30cm). The child was allowed five practise trials where the administrator would correct any inappropriate attempts (e.g., sliding the beanbag along the floor). This was followed by 10 formal trials where no feedback from the administrator was allowed. The score was the number of correct throwing attempts where the beanbag touched any part of the target circle during the formal trials (maximum raw score = 10).

5.2.3.5.5. Balance. Static balance was measured by the One Board Balance test from the M-ABC 2 (Henderson et al., 2007) whereby children were asked to balance with one foot on a plastic board with a thin keel. After a short practise, the child balanced with one foot on the board. Once the child had achieved a balanced position, the administrator began timing and continued for up to 30 seconds or when balance was lost. A maximum of two attempts of balancing for up to 30 seconds were allowed per foot. Raw scores were recorded for the duration of the longest overall balance time (maximum raw score = 30 seconds).

5.2.3.6. Executive Function. Memory was measured in the verbal and visual domains. Verbal memory was measured using the Digit Span task from the WISC-IV (Wechsler, 2004) and visual span was measured using the Block Recall task from the Working Memory Test Battery for Children (WMTB-C, Gathercole & Pickering, 2001). In addition, selective attention was measured using tasks from the Test of Everyday Attention for Children (TEA-Ch, Manly et al., 1998).

5.2.3.6.1. Verbal Span. The forward and backward digit span subtests from the WISC-IV (Wechsler, 2004) were used to measure verbal memory. In the forward subtest, the child was asked to recall sequences of single digit numbers the administrator read aloud. In the backward subtest, the child was asked to recall the single digit numbers in the reverse order. The sequence length increased from two to nine digits and the child recalled two trials per sequence length. Administration was discontinued when the child was unable to recall two trials of the same string length. The scores from the forward and the backward subtests to produce an overall score of verbal span (maximum score = 32)

5.2.3.6.2. Visual Span. Visual memory was measured using the Block Recall subtest of the WMTB-C (Gathercole & Pickering, 2001). During the test, the child tapped the same sequence of blocks as was demonstrated by the administrator. The span of blocks in the sequence increased from one to nine. Each span had a total of six trials and one point was

awarded per correct trial. The test was stopped when the child made three errors in one span. The number of correct trials prior to the discontinue rule was recorded (maximum score = 54).

5.2.3.6.3. Sky Search. Selective attention was measured using the Sky Search subtest. In the first block, the child was presented with an A3 sheet of paper with an array of 10 x 13 pairs of space ships. They were instructed to circle all the pairs of space ships where both space ships were the same (target pairs) whilst ignoring pairs of space ships that consisted of different ship designs (distractor ships). The child was encouraged to find and circle as many of the target pairs as quickly as s/he could and to tick a box located the bottom of the paper to signal they had finished searching. In a second, motor control, block the child was presented with another A3 sheet which only had the target pairs printed on it. The child was instructed to circle every item (all the pairs of ships) on the page see as quickly as possible. Scoring of the task followed the published instructions. The raw score was the time per target (the time taken to complete the task divided by the number of correctly circled target pairs) minus the time per target of the motor control block (the time taken to complete the task divided by the number of correctly circled target pairs).

5.2.3.6.4. Sky Search DT. Sky Search DT was used as a secondary measure of selective attention. In this task, the child was presented with a new 10 x 13 array of space ships printed onto A3 paper and was asked to circle as many target pairs as possible. At the same time, the child counted the number of 'scoring' sounds played via the tape and recall the number of sounds they heard when prompted by the administrator at the end of each block. The trial ended when the child indicated they had found all the space ships by ticking a box at the bottom of the paper. The raw score used here was the time per target as described for Sky Search.

5.2.4. Data Analysis

Due to the diverse age range in the sample, correlations were initially run to examine the relationship between age and performance on the tests. Correlations further examined the relationships between tests measuring the same and different skill domains. As multiple measures of phonological speed, literacy, perceptuomotor ability, motor skills, and executive functions were administered, the structure of the battery was examined using factor analyses. As in Study 1, exploratory factor analyses (EFA) and a confirmatory factor analysis (CFA) were run. To deal with age effects, a multiple indicators, multiple causes (MIMIC) model was specified by regressing the latent variables onto age. In using this approach, it was assumed that all children in the sample were part of a larger continuum whereby children with a disorder (e.g., dyslexia and/or DCD) would score at the tail end of these distributions on tests in which they were impaired.

To identify independent and shared markers of the disorders, factor score estimates were derived from the MIMIC model and were compared using a double dissociation 2 x 2 design with dyslexia status (present vs. absent) and DCD status (present vs. absent) as the independent variables. This two-way double dissociation design has been used in previous studies examining the cross over between neurodevelopmental disorders (e.g., Gooch et al., 2011; de Jong et al., 2006). Significant main effects would signal impairments and therefore markers of either dyslexia and/or DCD.

5.3. Results

A total of seven children were removed from these analyses, due to poor comprehension of the tasks ($n = 2$), very low global ability ($n = 1$), poor attendance ($n =1$), very high Welsh literacy scores and very low English literacy scores ($n = 2$), and a referred child not meeting any criteria for a disorder ($n = 1$).

5.3.1. Descriptive Statistics

The mean, standard deviation, range, and reliability for each measure is reported in Table 5.3. Composite measures of RAN Letters and Digits (RAN in Table 5.3) and One Minute Word and Nonword reading (Reading Fluency in Table 5.3) were derived to improve the reliability of the measures. Extreme outliers were Winsorized to 10th and 90th centile of the distribution. Overall, the distributions of the measures were acceptable (skewness < 1). Where possible reliabilities were calculated on a representative sample of unselected participants, normative samples. Generally, the measures ranged from having acceptable to excellent reliability. However, the measure of gross motor skill, Bag Throw, had poor internal consistency.

Table 5.3.

Descriptive Statistics of Measures for the Pooled Sample of Participants

	M	*SD*	Min	Max	Reliability
Phonological Speed					
Phoneme Awareness	118.39	36.50	73.7	234.5	.76[a]
RAN[†]	21.21	4.15	15.9	33.2	.84[a]
Literacy					
Word Spelling	25.28	6.27	10	40	.90[b]
Sentence Spelling	35.15	12.86	2	59	.94[b]
Word Reading	38.68	8.91	18	58	.94[b]
Reading Fluency[†]	63.58	17.34	20.5	105.5	.80[b]
Visuospatial					
Visual Perception	23.85	2.11	18	27	.64[b]
Visual Motor Integration	19.81	3.10	12	27	.73[b]
Motor					
Shape Tracing	21.71	2.57	16	26	.70[b]
Trail Drawing	1.25	1.27	0	5	.56[a]
Threading	25.71	10.07	16	111	.48[a]
Bag Throw	6.02	2.02	1	10	.48[b]
Balance	21.73	8.54	2	30	.73[a]
Executive Function					
Verbal Span					.49[a]
Visuospatial Span	25.90	4.22	9	38	.76[b]
Sky Search	4.20	1.29	0.3	8.0	.67[a]
DT Time per Target	5.87	2.01	2.9	13.9	.53[a]

Note. *N = 151*
[†]Composite measures. [a]Pearson's correlation. [b]Cronbach's alpha.

5.3.2. Relationships Within and Between Measures of Different Skill Domains

Pearson's correlations were run between all tests across the whole sample to examine the relationships between tests tapping the same and different domains (see Table 5.4). The strength of relationships between measures of similar and different constructs were examined to establish convergent and divergent validity. Large correlations between measures tapping the same construct demonstrated convergent validity whereas weaker correlations between measures tapping different constructs demonstrated divergent validity.

5.3.2.1. Phonological speed. There were moderate negative correlations between both the Phoneme Deletion and RAN measures and age meaning that older children were faster on both measures. There was also a strong correlation between both phonological speed measures suggesting good convergent validity. Phonological speed measures correlated highly with literacy measures, particularly with Reading Fluency. This was expected because the Reading Fluency measure – a composite of One Minute Word and One Minute Nonword Reading – draws heavily on phonological skills. Overall, smaller correlations were found between the phonological speed measures and perceptuomotor, motor, and executive function measures.

5.3.2.2. Literacy. There were also moderate correlations between age and literacy measures, meaning that reading and spelling scores increased with age. Large correlations between the reading and spelling measures indicated convergent validity. Moderate correlations with motor, memory, and attention measures indicated partial divergent validity.

5.3.2.3. Visuospatial. Small but significant correlations between age and the visuospatial measures were observed. Within the measures, there was a moderate correlation between Visual Motor Integration and Visual Perception suggesting that the tasks were measuring separable but related processes. There were no strong correlations with other measures suggesting some divergent validity. However, both measures had weak to moderate associations with measures from other domains, particularly motor and executive functions suggesting some association between visuospatial processes and other domains.

5.3.2.4. Motor. There was a poor relationship between age and functional motor skills, suggesting motor skill performance did not increase with age. Small to moderate correlations were found between measures of motor skills with the larger moderate correlations found on similar motor action measures. For example, Shape Tracing and Trail Drawing are both fine/graphomotor motor skill tasks. These associations reflect that these

measures tap a broad range of motor skills which differ in the use of fine and gross motor skills. Generally, the correlations between motor tests and measures tapping different constructs, such as executive functions, had smaller correlations demonstrating divergent validity. However, a measure of gross motor coordination, Bag Throw, had stronger correlations with literacy and phonological speed measures, than with other motor measures. This was unexpected, but correlations have been previously found between gross motor skills and reading ability (Lopes et al., 2013; Son & Meisels, 2006).

5.3.2.5. Executive Function. Performance on executive function, particularly on attention, moderately increased with age. Moderate correlations were also found between memory measures indicating partial convergent validity and, overall, there were smaller correlations between memory measures and tests tapping different domains (e.g., phonological speed), suggesting divergent validity. Visual span did correlate moderately with tests of motor skills. This correlation was expected as visual span taps motor planning and action processes. There were also moderate correlations between the memory and selective attention measures suggesting these measures were tapping separable but related mechanisms. On the selective attention measures, both correlated highly with each other suggesting good convergent validity. Together, executive function measures had weak to moderate relationships with measures of phonological speed, literacy, visuospatial, and motor measures underscoring that executive functions are tapped in most activities (Diamond, 2013). On balance, these associations were stronger than the associations between visuospatial tests and measures from other domains.

Table 5.4.

Correlations among Measures of Phonological Speed, Literacy, Visuospatial, Motor Skill, and Executive Functions

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17
1. Age	-																
Phonological Speed																	
2. Phoneme Deletion	-0.27***	-															
3. RAN	-0.32***	.68***	-														
Literacy																	
4. Reading Fluency	.32***	-.66***	-.83***	-													
5. Word Reading	.32***	-.57***	-.70***	-.74***	-												
6. Word Spelling	.36***	-.60***	-.64***	.70***	.85***	-											
7. Sentence Spelling	.36***	-.68***	-.69***	.74***	.87***	.92***	-										
Visuospatial																	
8. Visual Motor Integration	.22**	-.23***	-.18*	.21*	.25**	.29***	.31***	-									
9. Visual Perception	.24**	-.23**	-.20*	.16	.33***	.29***	.35***	.43***	-								
Motor																	
10. Motor Coordination	.21*	-.16*	-.17*	.13	.19*	.19*	.22**	.47***	.35***	-							
11. Trail Drawing	-.04	.23**	.16*	-.17*	-.21*	-.17*	-.21*	-.35***	-.16*	-.37***	-						
12. Threading	-.11	.18*	.20*	-.19*	-.1	-.1	-.21*	-.31***	-.25**	-.28***	.06	-					
13. Bag Throw	.13	-.33***	-.43***	.38***	.38***	.39***	.42***	.21**	.22**	.28***	-.33***	-.17*	-				
14. Balance	.20*	-.32***	-.32***	.21**	.25**	.23**	.24**	.22**	.22**	.32***	-.26**	-.19*	.18*	-			
Executive Function																	
15. Verbal Span	.17*	-.33***	-.31***	.24**	.35***	.36***	.35***	.29***	.37***	.22**	-.23**	-.01	.23**	.25**	-		
16. Visual Span	.21**	-.34***	-.32***	.30***	.23**	.30***	.31***	.36***	.34***	.33***	-.38***	-.23**	.36***	.36***	.39***	-	
17. Sky Search	-.32***	.48***	.51***	-.48***	-.41***	-.44***	-.49***	-.20*	-.18*	-.24**	.05	.24**	-.28***	-.20**	-.27***	-.24**	-
18. Sky Search DT	-.34***	.43***	.44***	-.37***	-.33***	-.41***	-.44***	-.36***	-.24**	-.26**	.15	.31***	-.24**	-.31***	-.26**	-.29***	.62***

Note. N = 151.

*p < .05, ** *p* < .01, ***p < .001.

5.3.3. Factor Structure of the Battery

Factor analyses techniques were used to investigate the factor structure of the battery. As in Study 1, exploratory factor analysis (EFA) was used to test the hypothesised five factor (phonological speed, literacy, visuospatial, motor, and executive function) structure. A baseline confirmatory factor analysis (CFA) was established to validate the structure of the battery. Due to the large variability in age, and that age was significantly associated with performance on most measures (see correlations), MIMIC modelling was used where the latent variables were regressed onto age.

5.3.3.1. Exploratory factor analysis (EFA). EFAs were run across the entire sample to test the hypothesised five factor structure. Maximum likelihood (ML) estimation was used to assess the goodness-of-fit indices between four-, five-, and six-factor solutions. Geomin rotation was used because correlations were expected. As in Study 1, Hu and Bentler's (1999) guidelines were used to assess the model fit in all factor analysis models. In brief, the guidelines suggest a good fitting model should have, as a minimum: χ^2 with $p > .05$, RMSEA < .06, SRMR < .08, CFI and TLI > .95. Table 5.5 shows that the four, five, and six factor solutions all provided an acceptable fit.

Table 5.5.

Goodness-of-Fit and Chi Square Difference Tests for Exploratory Factor Analyses of

Measures Assessing Phonological Speed, Literacy, Visuospatial, Motor, and Executive Skills

Solution	χ^2	df	RMSEA	SRMR	CFI	TLI	χ^2_{diff}	Δ_{df}
4-Factor	89.06^{ns}	74	.04	.03	0.99	.98	-	-
5-Factor	67.70^{ns}	61	.03	.02	1.00	.99	$21.36^{ns\dagger}$	61
6-Factor	40.57^{ns}	49	.00	.02	1.00	1.02	$27.13^{**\ddagger}$	49

Note. $N = 151$.
[†]Chi Square difference between four and five factor solutions. [‡]Chi Square difference between five and six factor solutions. RMSEA, root mean square of error approximation; SRMR, standardised root mean square residual; CFI, comparative fit index; TLI, Tucker-Lewis index.
p < .05, ** *p* < .01, ***p* < .001

The factor loadings and the communality of the four- (Table 5.6) and five-factor (Table 5.7) solutions were acceptable in the main. In both solutions, reading and spelling loaded onto one factor along with phonological speed measures. In addition, the phonological speed measures loaded onto a second factor. Also, in both models, the measures of visuospatial and motor skill loaded onto the same factor suggesting that these measures were tapping the same underlying perceptuomotor construct. In the four-factor solution the memory measures loaded onto the same factor as the motor measures but in the five-factor solution, memory measures loaded onto a separate factor. The selective attention measures did not load consistently well onto any factor in either model.

Table 5.6.

Factor Loadings and Communality Values for the Four-Factor Exploratory Factor Analysis of Measures Assessing Phonological Speed, Literacy, Visuospatial, Motor, and Executive Skills

	Factors				Communality
	1	2	3	4	
Phoneme Deletion	**-.55***	**.28***	-.10	-.09	.57
RAN	**-.59***	**.59***	-.01	.00	.87
Reading Fluency	**.68***	**-.45***	-.03	-.00	.79
Word Reading	**.90***	-.04	.03	-.10	.83
Word Spelling	**.96***	.07	.01	-.02	.90
Sentence Spelling	**.97***	.05	-.01	.10	.96
Visual Motor Integration	.05	.14	**.53***	**.30**	.50
Visual Perception	.12	.13	**.44***	.19	.35
Shape Tracing	-.06	.01	**.58***	**.28**	.50
Trail Drawing	.07	.04	**-.68***	.07	.41
Threading	.01	.14	-.02	**-.63***	.44
Bag Throw	**.22***	**-.20**	**.34***	-.02	.31
Balance	.02	**-.21***	**.39***	.06	.25
Verbal Span	**.22***	.01	**.49***	-.16	.33
Visual Span	.01	-.14	**.60***	.03	.43
Sky Search	-.04	-.17	.16	-.01	.06
Sky Search DT	**.33***	-.12	.01	**.37**	.34

Note. Factor loadings > .2 are in boldface.
*$p < .05$.

Table 5.7.

Factor Loadings and Communality Values for the Five-Factor Exploratory Factor Analysis

of Measures Assessing Phonological Speed, Literacy, Visuospatial, Motor, and Executive

Skills

	Factors					Communality
	1	2	3	4	5	
Phoneme Deletion	**-.55***	**.24***	-.06	-.12	-.12	.58
RAN	**-.65***	**.54***	.01	-.02	-.05	.87
Reading Fluency	**.75***	**-.41***	.01	-.05	-.01	.80
Word Reading	**.93***	-.02	-.01	.03	-.13	.84
Word Spelling	**.97***	.09	-.03	.01	-.02	.90
Sentence Spelling	**.97***	.09	.03	-.02	.04	.95
Visual Motor Integration	.03	**.22***	**.64***	.04	.09	.50
Visual Perception	.06	.19	**.37***	**.22**	.13	.36
Shape Tracing	-.05	.09	**.76***	-.03	.02	.52
Trail Drawing	.00	.01	**-.83***	-.01	**.45***	.58
Threading	-.01	.03	**-.41***	**.25**	**-.36***	.38
Bag Throw	**.28***	-.16	**.39***	-.01	-.17	.33
Balance	.03	-.16	**.36***	.11	.00	.24
Verbal Span	.04	.00	.02	**.79***	-.01	.67
Visual Span	-.02	-.09	**.46***	**.28**	-.01	.42
Sky Search	-.07	-.15	.05	.18	.09	.07
Sky Search DT	**.28***	-.03	.05	.05	**.46***	.42

Note. Factor loadings > .2 are in boldface.
*p < .05.

5.3.3.1. Confirmatory factor analyses. Confirmatory factor analyses (CFA) were

run to validate the factor structure of the battery. Initially, a baseline measurement model was

established amongst the whole sample. Age was then added to the model to account for any

variance that could be explained by age.

5.3.3.1.1. Baseline CFA. CFAs were run using direct ML estimation in Mplus 7.2

(Muthén & Muthén, 2014) due to the small amount of missing data completely at random

(9% on Reading Fluency and Threading, 7% on Trail Drawing, and < 2% across all other

measures). All indicators were entered the model. All indicators were loaded onto their

hypothesised constructs (see Table 5.2) except for the visuospatial and motor measures.

Because visuospatial and motor measures consistently loaded onto the same factor in the

EFAs, they were combined to produce a perceptuomotor factor in the model. The indicator, Bag Throw, had high standardised residuals (> 2.50) and modification indices (> 4.00) suggesting an area of localised strain. Taken together with the fact that gross motor skills were the least likely to be impaired in children with dyslexia (Iversen et al., 2005), Bag Throw was removed from the model. Removing Bag Throw improved the overall fit. Further iterations of model development involved freeing the cross loadings and correlating residuals of indicators where there was substantial theoretical justification for doing so.

Reading Fluency was cross-loaded onto phonological speed because this measure taps phonological skills. Residuals were also correlated between literacy and language measures. The residuals of RAN and the One Minute Reading composite were correlated to account for the variance shared by rapid word production. Phoneme Deletion was correlated with sentence spelling, an association that has been reported previously (see Caravolas et al., 2005). Finally, Sky Search and Sky Search DT were correlated to account for the variance shared by similar task demands. The final baseline model produced an acceptable fit, $\chi^2(94)$ = 142.35, p = .001, RMSEA = .06 [90% C.I. = .04, .08], SRMR = .06, CFI = .96, and TLI = .96 with no large modification indices, indicating no areas of strain. In this model (see Figure 5.2) all indicators significantly loaded onto their respective constructs. There were strong correlations between the phonological speed and literacy latent variables. There were also large correlations between executive function and all other latent variables. Phonological speed and literacy both moderately correlated with the motor factor.
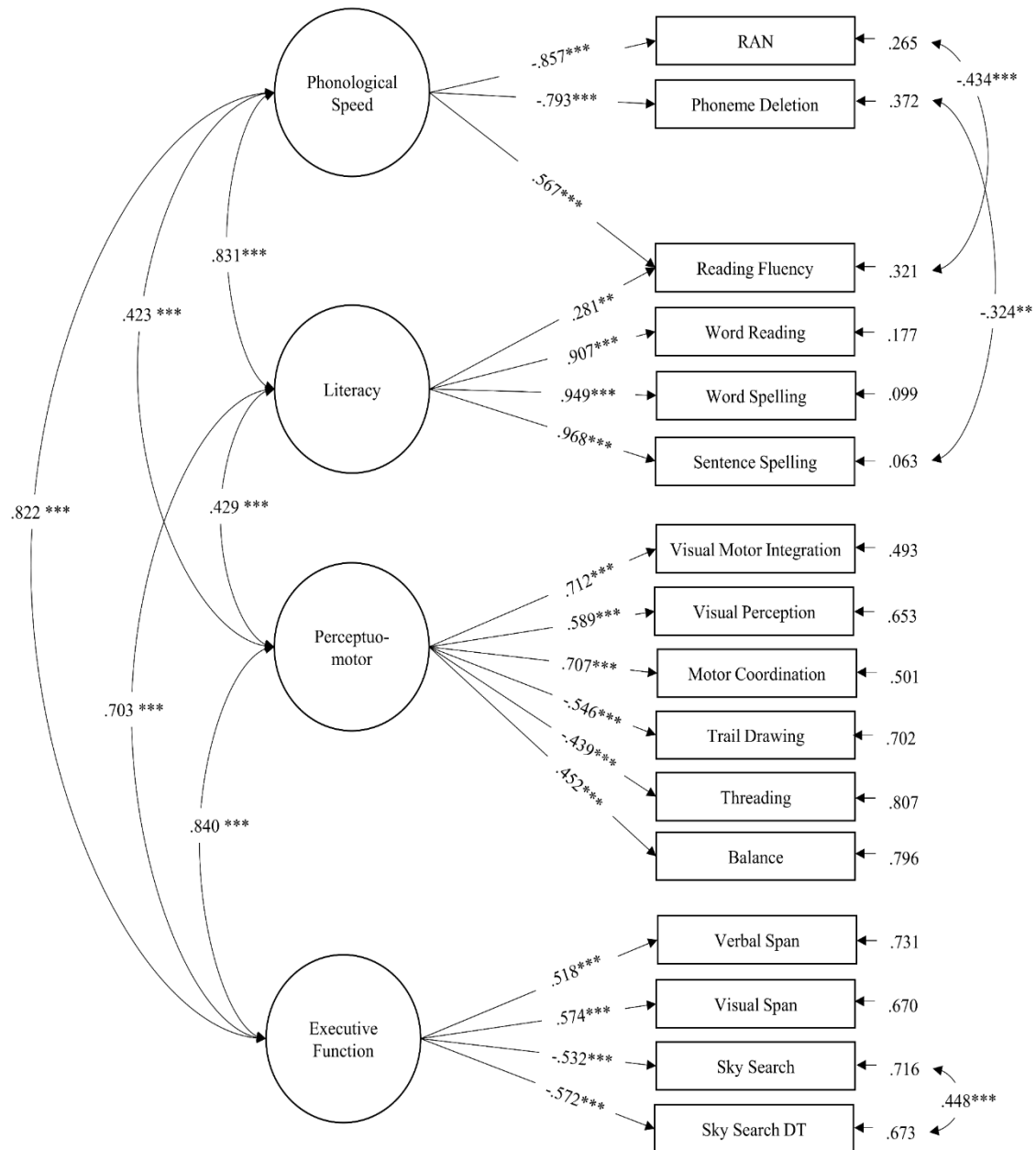
Figure 5.2. Path model of marker battery baseline CFA. The path model includes correlations, standardised factor loadings, and residual variances of the measures loaded onto the four latent variables, phonological speed, literacy, motor, and executive function. *p < .05, ** p < .01, ***p < .001.

5.3.3.1.2. MIMIC Model. Age was added as a covariate into the baseline model to account the effects of age on the latent variables. A MIMIC model was favoured over a multi-group confirmatory factor analysis (MGCFA) used in the previous study due to the

relatively small sample size ($N = 151$) and the extended data collection period for this study (see General Methods), precluding the use of class year as a grouping variable. The four latent variables were regressed onto the age covariate. No indicators were regressed onto age as there were no specific predictions regarding direct effects of age on any specific indicator. The residuals between latent variables were also correlated because age alone cannot fully account for overlap between the constructs.

The resulting MIMIC model produced an acceptable fit, $\chi^2(106) = 153.53$, $p = .002$, RMSEA = .05 [90% C.I. = .034, .072], SRMR = .06, CFI = .97, and TLI = .96, with significant loadings of all indicators onto their respective constructs (see Figure 5.3). The inclusion of age into the model did not alter the factor structure or introduce new areas of strain into the model (e.g., there were no different modification indices between the baseline and MIMIC models). The regression paths between age and the latent variables were all small to moderate but significant and the direction of these parameter estimates indicates that performance on each of the latent constructs improved with age. As in the baseline model, large significant correlations were found between the latent variables of literacy and phonological speed. Executive function produced large correlations all other latent variables. There were moderate correlations between motor skills and literacy and phonological speed.
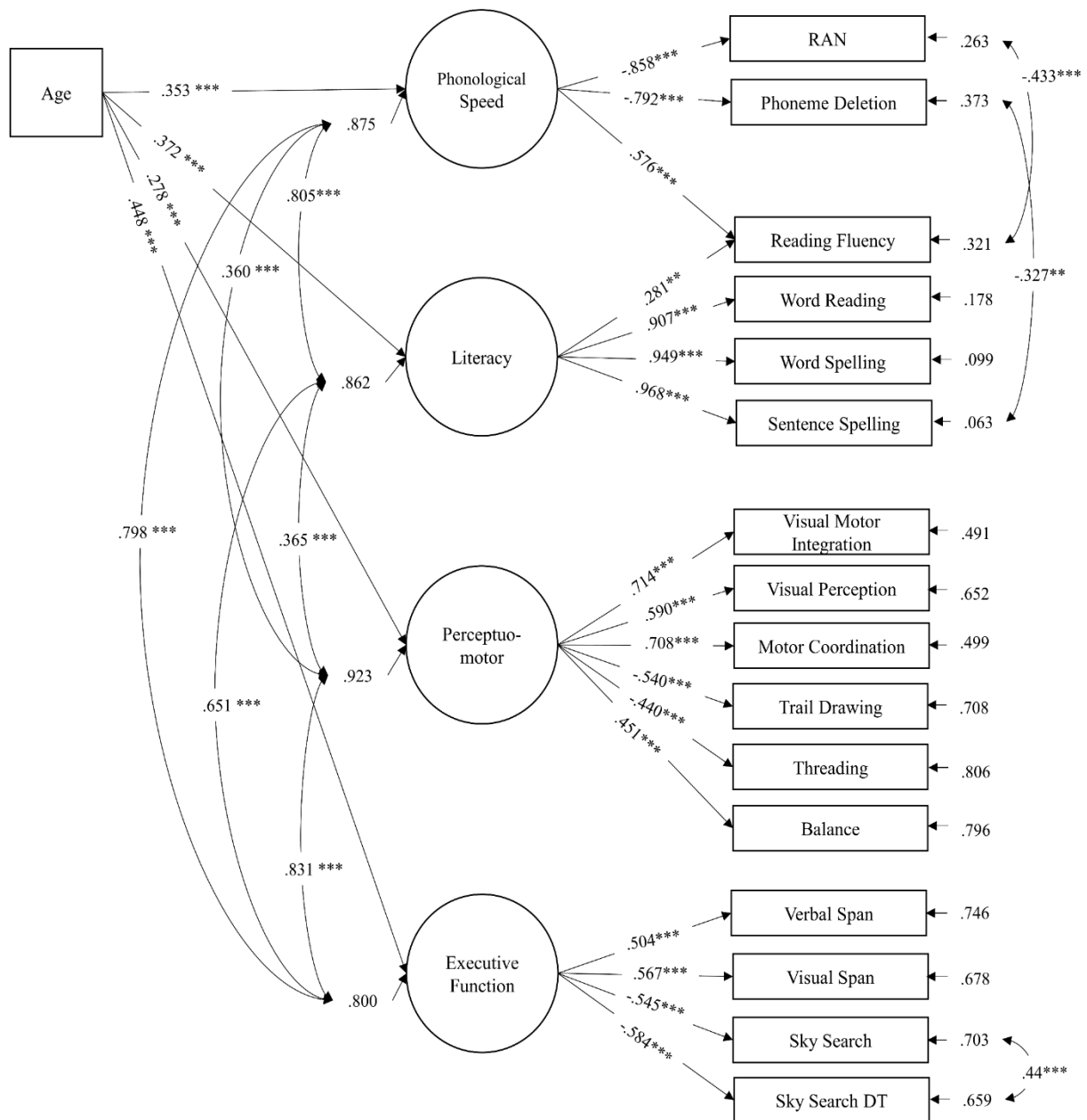
Figure 5.3. Path model of marker battery MIMIC model including an age regressor (*N* =

151). Standardised parameter estimates, residual variances, and correlations between latent

variables are reported. Performance on all latent variables significantly increased with age.

p < .05, ** *p* < .01, ***p* < .001.

5.3.3.2. Deriving factor score estimates. Following the validation of the factor

structure of the battery, refined factor score estimates were calculated from the model in

Mplus using the regression approach. Factor score estimates act as proxy measures of each

participant's relative position on the respective latent variables and provide a 'purer' measure of the latent variable by removing the variability which would be associated with analyses of each individual test (Brown, 2015). A major issue concerning the use of factor score estimates, however, is their indeterminacy. This means that any number of factor score estimates could be calculated from the model and still be consistent with the model's factor loadings (Brown, 2015; Grice, 2001).

The degree of indeterminacy can be assessed using several criteria (Grice, 2001). In this study, the quality of the factor score estimates derived from the current model was assessed using validity coefficients and correlational accuracy. Validity coefficients are the correlations between the factor score estimates and their respective factors (Grice, 2001). Grice (2001) recommends the validity coefficients should be > .8 and ideally > .9 if factor score estimates are to be used as dependent variables. As Table 5.8 reports, all values were > .9 indicating these scores were suitable for use as dependent variables.

Table 5.8.

Validity Coefficients of Phonological Speed, Literacy, Motor Skills, and Executive Function Factor Score Estimates

Factor	Validity Coefficient
Phonological Speed	.94
Literacy	.98
Perceptuomotor	.90
Executive Function	.94

The factor score estimates were also assessed for correlational accuracy which is the degree to which the correlations among the factor score estimates corroborate the correlations among the factors in the model (Grice, 2001). The correlations among the extracted factor score estimates were greater in magnitude than the correlations reported between the factors (see Table 5.9). However, the differences between the matrices were small, with the largest

difference between phonological speed and motor skills ($r = .15$). In sum, the quality of the

factor score estimates was acceptable and therefore were used in the following analyses to

compare the profiles of children with dyslexia, DCD, and comorbid dyslexia and DCD.

Table 5.9

Correlations between Extracted Refined Factor Score Estimates (top portion) and Model

Factor Scores (bottom portion)

	1	2	3	4
1. Phonological Speed	-	.89	.51	.86
2. Literacy	.81	-	.49	.77
3. Perceptuomotor	.36	.37	-	.87
4. Executive Function	.80	.65	.83	-

Note. All correlations $p < .001$.

5.3.4. Profiles of Ability in Dyslexia and/or DCD

Profiles of phonological speed, literacy, perceptuomotor, and executive function

abilities were examined to identify independent and shared markers of dyslexia and DCD. To

do so, 2 (dyslexia: present vs absent) x 2 (DCD: present vs absent) weighted ANCOVAs

were run on each of the four – phonological speed, literacy, perceptuomotor, and executive

function – factor score estimates using Stata 13 (Stata Corp., 2013). Weighted analyses were

used due to the large differences in sample sizes between the groups (see Table 5.1). In all

models, Block Design was initially entered as a covariate to control for group differences in

NVIQ. In models where NVIQ was a non-significant covariate (phonological speed and

literacy), the models were re-run without the covariate and are reported instead of the

ANCOVAs.

The presence of a significant main effect of dyslexia indicate that children with

dyslexia received lower factor score estimates than children without dyslexia. This would be

suggestive of impaired performance amongst children with dyslexia. Similarly, the presence

of a significant main effect of DCD would indicate that children with DCD received lower

factor score estimates than children without DCD which would suggest impaired performance amongst children with DCD. Following the AN(C)OVAs, singular and comorbid groups were compared using pairwise comparisons with Bonferroni corrections to elucidate the profiles of comorbid dyslexia and DCD relative to dyslexia-only and DCD-only groups. The means, standard deviations, 2 x 2 AN(C)OVAs, and follow up comparisons for each factor are reported in Table 5.10. The results of these analyses are now discussed for each factor separately.

5.3.4.1. Phonological Speed. Children with dyslexia (singular and comorbid) achieved lower factor score estimates than children with DCD-only and typically developing children. The assumption of homogeneity of variance was broken and so the data were transformed by squaring each score. This improved the distribution and the assumption of homogeneity of variance was no longer broken. In the initial 2 x 2 ANCOVA, the covariate of NVIQ was non-significant and the analysis was re-run without NVIQ. The resulting ANOVA revealed a large effect of dyslexia, a non-significant effect of DCD and a non-significant interaction (see Table 5.10). Pairwise comparisons between singular and comorbid groups revealed both dyslexia groups had significantly lower factor score estimates than typically developing children and children with DCD ($ps < .001$). The dyslexia groups did not differ significantly, nor did children with DCD differ significantly from typically developing controls.

4.3.4.2. Literacy. On the literacy factor, children with dyslexia (singular and comorbid) achieved scores similar in magnitude that were smaller than children with DCD and typically developing controls. In this analysis, NVIQ was a nonsignificant covariate and so was removed. The 2 x 2 ANOVA revealed a significant main effect of dyslexia, but no significant effect of DCD or interaction between the disorders. Follow-up comparisons confirmed both dyslexia groups had significantly lower factor score estimates than typically

developing children and children with DCD (*ps* < .001). The dyslexia groups (singular and comorbid) did not differ from each other.

5.3.4.3. Perceptuomotor. Children with singular and comorbid DCD had lower perceptuomotor scores than typically developing children and children with dyslexia-only. After controlling for NVIQ, there was a moderate effect of DCD status, but no significant effect of dyslexia or interaction between disorders. Post-hoc comparisons confirmed that children with DCD (singular and comorbid) had significantly lower motor scores than typically developing children and children with dyslexia (*ps* < .01), but children with DCD-only and comorbid dyslexia and DCD did not differ significantly from one another.

As noted earlier, during the factor analyses it became clear that the visuospatial and the functional motor measures loaded onto the same, perceptuomotor, factor. To further examine performance on measures of visuospatial abilities in dyslexia and DCD, two further 2 x 2 ANCOVAs were performed on the standardised scores on the Visual Perception and Visual Motor Integration tests in addition to the analysis of the perceptuomotor factor score estimates discussed above.

4.3.4.3.1. Visual perception. On average, all groups performed within the average range for their age. Typically developing children (*M* = 100.91, *SD* = 8.45), children with dyslexia-only (*M* = 99.46, *SD* = 7.68), and children with DCD (*M* = 97.08, *SD* = 10.02) scored very similarly to one another and children with comorbid dyslexia and DCD achieved slightly lower scores (*M* = 91.12, *SD* = 11.9). Although groups performed similarly on average, there was more variance in the singular and comorbid DCD groups than in the typically developing and dyslexia-only groups. The 2 x 2 ANCOVA of visual perception scores revealed no significant main effects of dyslexia, $F(1, 134) = 1.21$, $p = .273$, $\eta_\rho^2 = .01$, or DCD, $F(1, 134) = 1.71$, $p = .193$, $\eta_\rho^2 = .01$, and no significant interaction between them, $F(1, 134) = 0.25$, $p = .620$, $\eta_\rho^2 < .01$.

5.3.4.3.2. Visual motor integration. Children with DCD-only ($M = 80.84$, $SE = 12.6$) and children with comorbid dyslexia and DCD ($M = 74.53$, $SE = 9.96$) performed less well than typically developing children ($M = 94.1$, $SD = 8.97$) and children with dyslexia-only ($M = 90.64$, $SE = 12.59$). Accordingly, there was moderate effect of DCD, $F(1, 134) = 15.85$, $p < .001$, $\eta_\rho^2 = .12$, but no significant effect of dyslexia, $F(1, 134) = 2.35$, $p = .128$ $= .02$, or interaction, $F(1, 134) = 0.16$, $p = .694$, $\eta_\rho^2 < .01$. Follow-up pairwise comparisons revealed children with DCD-only and comorbid dyslexia and DCD achieved significantly lower visual-motor integration scores than typically developing children ($ps < .01$). No other pairwise comparisons reached statistical significance.

5.3.4.4. Executive function. All disorder groups performed less well than typically developing children. Of the disorder groups, children with singular dyslexia and DCD performed similarly and children with comorbid dyslexia and DCD achieved the lowest average scores. There were moderate effects of dyslexia and DCD status, but no significant interaction between the disorders. Follow-up comparisons confirmed that all three disorder groups had significantly lower executive function factor score estimates than typically developing children ($ps < .01$). Factor score estimates did not differ significantly between children with dyslexia-only and DCD-only ($p > .999$). The factor score estimates of children with comorbid dyslexia and DCD were significantly lower than both dyslexia-only ($p = .028$) and DCD-only groups ($p = .014$). However, it is important to note that the lack of significant interaction between dyslexia and DCD status suggests that the comorbid profile was an additive combination of deficits of dyslexia and DCD.

Table 5.10.

Means, Standard Deviations, Main Effects, Covariates, and Interactions from the 2 x 2 Weighted AN(C)OVAs of Factor Score Estimates

| | DYS[a] | | DCD[b] | | DYS+DCD[c] | | TD[d] | | Main Effect | | | | Covariate | | Dyslexia x DCD | |
| | | | | | | | | | Dyslexia | | DCD | | NVIQ | | | |
	M	SD	M	SD	M	SD	M	SD	F	η_ρ^2	F	η_ρ^2	F	η_ρ^2	F	η_ρ^2
Phonological Speed	1.70[bd]	.4	2.36	.35	1.39[bd]	.48	2.46	.32	48.25***	.25	2.10	.01	-	-	0.19	<.01
Literacy	2.89[bd]	.49	4.10	.80	2.70[bd]	.51	4.32	.7	46.77***	.24	0.95	.01	-	-	0.01	< .01
Perceptuomotor	2.31	.37	1.7[ad]	.41	1.32[ad]	.48	2.45	.47	3.65	.03	38.42***	.22	18.99***	.12	0.76	.12
Executive Function	2.27[d]	.35	2.33[d]	.28	1.73[abc]	.39	2.69	.34	22.27***	.14	16.80**	.11	4.43*	.03	0.66	< .01

Note. DYS = dyslexia. DYS+DCD = comorbid dyslexia and DCD. TD = typically developing. NVIQ = non-verbal IQ. The covariate, NVIQ, is not reported for phonological speed and literacy analyses as NVIQ was a non-significant covariate and so was dropped from the model. Subscript of means represent significant post-hoc tests with Bonferroni corrections.
*p < .05, ** p < .01, ***p < .001.

5.4. Discussion

This study aimed to investigate the independent and shared markers between dyslexia and DCD and to examine the nature of comorbidity between the two disorders. These aims were addressed by comparing groups using factor score estimates extracted from a latent model. Factor score estimates are proxy measures of each participant's relative position on a latent variable and so provide a 'purer' measure than individual tests. As expected, children with dyslexia had deficits in phonological speed and literacy skills but unimpaired perceptuomotor abilities. Conversely, children with DCD had deficits in perceptuomotor abilities (including visual-motor integration) but spared phonological speed and literacy skills. Deficits in executive function were apparent in both dyslexia and DCD. A second aim of this study was to examine the profiles of children with comorbid dyslexia and DCD relative to children with singular dyslexia and DCD. Children with comorbid dyslexia and DCD performed similarly to children with dyslexia-only and/or DCD-only in all domains. These findings are discussed in relation to the literature. Further issues also discussed are limiting condition and practical implications.

5.4.1. Dyslexia and DCD: Independent and Shared Markers

The multiple deficit model (MDM) conceptualises neurodevelopmental disorders at the aetiological, neural, cognitive, and symptom levels (Pennington, 2006). This study focused on the examining independent and shared markers of cognitive deficits and behavioural symptoms. The literatures on dyslexia and DCD suggest a high degree of overlap in these domains and a primary aim of this study was to directly compare dyslexia and DCD to elucidate shared and independent markers.

5.4.1.1. Phonological processing. As expected, children with dyslexia but not DCD had deficits in phonological speed. This is consistent with a large literature describing phonological processing deficits in dyslexia (Vellutino et al., 2004) but contradict others who

report phonological-related difficulties in children with DCD at the group level (Archibald & Alloway, 2008; Dewey et al., 2002). The reason for this disparity in findings with regards to children with DCD is most likely due to a lack of control of comorbid cases in previous studies. Notably, Dewey et al. (2002) did not discriminate between children with DCD and phonological problems who had reading and spelling difficulties (probable dyslexia) and those who did not. Thus, phonological processing deficits are a marker of dyslexia but not DCD.

      5.4.1.2. Executive functions. Previous studies had reported impairments in memory and attention in both dyslexia and DCD (Alloway, 2007; Cruddace & Riddell, 2006; Swanson et al., 2009), suggesting deficits in executive functions could constitute a shared risk factor or marker for dyslexia and DCD. Yet, potential confounds namely in measurement issues left the presence of shared deficits an open question. Many of the aforementioned measurement issues were overcome using a latent modelling approach. By examining the variance shared between these measures we were able to remove variance that might be explained by other aspects of the task and tap into the shared variance explained by executive function. When doing so, we found deficits in executive functions in both dyslexia and DCD. Such deficits in executive function deficits are presumably not causally related to dyslexia and DCD, rather they may compound difficulties which increases the likelihood of children meeting a diagnostic threshold (Gathercole et al., 2016; Hulme & Snowling, 2009).

      5.4.1.3. Perceptuomotor ability. In this study, we initially set out to examine visuospatial and functional motor skills as separate constructs. However, it became apparent that the measures of these constructs shared a large amount of variance and so were considered jointly under perceptuomotor ability. The large amount of shared variance between these measures highlights the close relationship between perceptual abilities (visuospatial ability) and functional motor skills. On this construct, children with DCD, but

not dyslexia, performed poorly. These findings are inconsistent with studies reporting

impaired perceptuomotor performance amongst children with dyslexia (e.g., Iversen et al.,

2005; Nicolson & Fawcett, 1990). This inconsistency is likely to reflect the inclusion of

children with comorbid dyslexia and DCD in previous studies (Rochelle & Talcott, 2006).

It is important to note that children with dyslexia did perform less well on the gross

motor selection measure, Bag Throw, implying there may have been some sub-clinical motor

impairments in dyslexia. However, this measure was not highly related to other motor

measures (it correlated more strongly with phonological and literacy measures), nor was the

internal consistency of the measure good, raising questions about the appropriateness of this

test. In addition, previous research has suggested gross motor skills were the least impaired

aspect of motor skills amongst dyslexics (e.g., Iversen et al., 2005). Therefore, for these

reasons it is unlikely that children with dyslexia had any subclinical impairments in (gross)

motor skills.

As mentioned previously, visuospatial measures (Visual Perception and Visual Motor

Integration) shared a large amount of variance with functional motor measures in the analyses

and were subsequently combined to produce a perceptuomotor factor. Subsequently, separate

analyses revealed children with DCD but not dyslexia had impairments in visual motor

integration. These findings are consistent with previous reports of deficits in visual motor

integration amongst children with DCD (e.g., Bonifacci, 2004, Schoemaker et al., 2001; Van

Waelvelde et al., 2004) and suggests deficits in visual motor integration are a marker of DCD

but not dyslexia.

Unexpectedly, children with DCD performed no differently to children without DCD

on a visual discrimination task, indexing visual perception. This finding implies children with

DCD did not have a visual perception deficit and contradicts several reports that find such

impairments (e.g., Hulme, Smart, & Moran, 1982; Tsai, Wilson, & Wu, 2008; Wilson et al.,

2013). The contradictory findings might be explained by differences in the way visual perception was measured. For example, Tsai et al. (2008) measured several aspects of visual perception in children with DCD and although children with DCD achieved significantly lower scores across all measures, only a small proportion of their DCD sample (28%) were impaired on a visual discrimination task analogous to the one administered here. Furthermore, visuoperceptual measures such as visual perception tests which have a smaller motor component, tend to discriminate between children with and without DCD less well than complex visuospatial tests with a motor component (Wilson et al., 2013). An additional issue was the visual perception test administered in this study had questionable internal consistency (see Table 5.3) which also raises questions about its utility as a measure of visuospatial skills.

    5.4.1.4. Literacy. Finally, we addressed potential literacy impairments amongst children with DCD. Previous studies had reported children with DCD had performed less well than typically developing children on reading and spelling tests, although performance was highly variable, and most studies had failed to screen for comorbid dyslexia (Alloway, 2007; Archibald & Alloway, 2008; Dewey et al., 2002; Schoemaker et al., 2013). There was no evidence of literacy impairments in children with DCD in this study, suggesting literacy impairments amongst children with DCD most likely to reflect cases of comorbidity in the sample.

5.4.2. Nature of Comorbidity between Dyslexia and DCD

    A second aim was to examine the profiles of children with comorbid dyslexia and DCD to test three competing hypotheses of the basis of comorbidity – phenocopy, cognitive subtype, shared aetiology – between the disorders. Children with comorbid dyslexia and DCD performed similarly to children with dyslexia-only and/or DCD-only in all domains. Notably, on executive function – impaired in both dyslexia-only and DCD-only groups –

children with comorbid dyslexia and DCD had larger deficits than the singular groups. Moll et al. (2016) found a similar pattern of larger deficits amongst children with comorbid dyslexia and dyscalculia when compared to children with singular disorders on measures of verbal memory. In both the current study and in Moll et al. (2016), the absence of a statistical interaction between dyslexia and the comorbid group suggests deficits in the comorbid group was an additive effect of deficits in both singular groups. Taken together, these findings are most consistent with a shared aetiology hypothesis (de Jong et al., 2006) and add to the growing evidence in favour of a shared aetiology between dyslexic heterotypic comorbidities (e.g., Gooch et al., 2011; Moll et al., 2016). A shared aetiology account proposes that the comorbid disorder results from shared genetic origins, which is consistent with the MDM's predictions of shared risk aetiological risk factors (Pennington, 2006).

5.4.3. Implications for Assessment and Intervention

The current findings have important implications for the way practitioners and researchers identify developmental disorders. Given the high rate of comorbidity between dyslexia and DCD (Study 1) practitioners and researchers should be encouraged to test for additional disorders. Furthermore, because comorbidity between dyslexia and DCD is additive, existing measures rather than new, comorbid disorder-specific measures, can be applied in combination to assess comorbidity. In the case of dyslexia and DCD, the current findings suggest a combination of literacy (standardised reading and spelling) and motor (M-ABC 2) tests would be adequate to identify potential comorbid dyslexia and DCD.

The current findings also have implications for designing interventions for children with comorbid difficulties. When designing interventions, it is important to always consider difficulties relating to the comorbid disorder. For example, when developing a targeted literacy intervention for a child with comorbid dyslexia and DCD, the amount of motor skill required by the child in completing the activity should be considered. Given that comorbidity

reflects an additive combination, further work should examine the efficacy of implementing existing literacy and motor skill interventions concurrently for children with comorbid dyslexia and DCD.

5.4.4. Considering Dyslexia and DCD in the Multiple Deficit Model: Revisited

This study focused on examining dyslexia and DCD within two aspects of the MDM. The first was to identify independent and shared markers of dyslexia and DCD. Phonological deficits and literacy impairments were found to be independent markers of dyslexia. Whereas perceptuomotor (and visual motor integration) deficits were independent markers of DCD. Both dyslexia and DCD shared markers in executive function deficits which could also partly explain the high degree of comorbidity between the disorders. The second aspect of the MDM focused on here was the nature of comorbidity between dyslexia and DCD. The profiles of children with comorbid dyslexia and DCD was best explained by the shared aetiology hypothesis. The shared aetiology hypothesis suggests that dyslexia and DCD share some common genetic aetiology. This hypothesis is closely aligned with the MDM which suggests that comorbid disorders result from some shared aetiological and cognitive risk factors Pennington, 2006). Thus, deficits in phonological and/or perceptuomotor processing along with deficits in executive functions and additional risk factors increase the risk of difficulties in developing literacy and/or (possibly) motor skills.

5.4.5. Limiting Conditions

Whilst this study has many implications, it is important to consider some potential limiting conditions, particularly in the way motor skills were examined. An aim of this study was to compare visuospatial ability and functional motor skills as separate constructs. Accordingly, tests were administered to assess different aspects of visuospatial ability and motor skills. However, during the analyses, it became apparent that tests of visuospatial ability and motor skills were tapping the same construct of perceptuomotor ability. On a

related note, during the analysis it also became apparent that our measure of gross motor skill was not tapping the same construct as the other motor measures and had to be removed from the model. Removing the only measure of gross motor skill meant that gross motor skill could not be compared between dyslexia and DCD in the present study. However, previous work has shown that gross motor skills were least likely to be impaired in children with dyslexia (e.g., Iversen et al., 2005). Both the issues with separating perceptuomotor skills from functional motor skills and the measurement of gross motor skill could reflect known problems with existing measures in adequately assessing motor skills (Gomez & Siringu, 2015; Venetsanou et al., 2011). One way to reduce the likelihood of problems relating to motor skill assessments would be to administer a larger range of measures hypothesised to be tapping the same aspects of visuospatial and functional motor skills.

5.4.6. Conclusion

This study was concerned with understanding the relationship between dyslexia and DCD. It focused on disentangling the apparent overlap between dyslexia and DCD and understanding the nature of comorbidity between the disorders. Once controlling for comorbidity, it was clear dyslexia and DCD were separable disorders with independent markers. Yet, both disorders share markers in executive functions. Children with comorbid dyslexia and DCD had deficits that were additive in nature suggesting a shared aetiology. Taken together, dyslexia and DCD are two neurodevelopmental disorders that result from independent and shared genetic and environmental risk factors acting to affect neural and cognitive development.

Returning to a key objective of this thesis which was to elucidate the nature of spelling and handwriting impairments in dyslexia and DCD. The finding in this study suggest that spelling impairments are present in dyslexia but not DCD, after controlling for comorbidity. This addresses the question about the nature of spelling impairments in dyslexia

and DCD. Yet, what remains unclear is the nature of handwriting difficulties in dyslexia

and/or DCD and their relationship with the impairments of the disorders found in this study.

The following studies are dedicated to this pursuit.

Chapter 6

Study 3: The Nature of Handwriting Legibility and Fluency Difficulties in Dyslexia and DCD

6.1. Introduction

Children with dyslexia and children with DCD have been described as having dysfluent and illegible handwriting (Berninger et al., 2008; Blank et al., 2012). Handwriting itself is a complex ability that involves the integration of literacy, motor, and cognitive skills. It follows that in Study 2 we found children with dyslexia and/or DCD were characterised by independent and shared impairments in skills that are related to handwriting. Some authors have linked impairments in these skills to handwriting difficulties in dyslexia and DCD. Specifically, handwriting fluency difficulties in dyslexia have been most strongly linked to literacy-related deficits (e.g., Sumner et al., 2013) whereas poor fluency amongst children with DCD have been associated with motor deficits (e.g., Prunty et al., 2013).

The current evidence, predominantly based on studies of handwriting fluency, implies that there are (a) dissociable profiles of handwriting difficulties between dyslexia and DCD and (b) handwriting difficulties have different bases in dyslexia and DCD. Yet, no study has directly compared handwriting profiles to assess whether there are dissociable differences in profiles of handwriting fluency and legibility. Furthermore, no study has examined the relationship between handwriting abilities and a wide range of skills related to handwriting in dyslexia and DCD. These were the main aims of the present study. In what follows, we describe how the abilities that were identified as marker impairments of dyslexia and/or DCD in Study 2 are related to handwriting. We then critically review the literature which describes handwriting difficulties in dyslexia and DCD and consider the current evidence of the associations between handwriting difficulties and handwriting related skills in dyslexia and DCD.

6.1.1. Skills Related to Handwriting

Although considered a lower level skill under the umbrella term of *transcription* (e.g., Abbott & Berninger, 1993; Berninger & Winn, 2006), handwriting is itself a complex ability

that involves the coordination and integration of several skills. Berninger and colleagues reported on the associations of orthographic and fine motor skills with handwriting (Abbott & Berninger, 1993; Berninger et al., 1992, 1994; Berninger & Rutberg, 1992). This work highlighted that handwriting is more than just a motor-based ability. It is one that is related to language/literacy abilities as well. Here, we build on the earlier work by Berninger and colleagues to consider how a broad range of skills may be related to handwriting ability in dyslexia and DCD. The psychomotor model (van Galen, 1991) provides a detailed account of the lower level sub process involved in handwriting (see Chapter 1, p. 14) and so it is a useful framework for mapping how associated skills are related to handwriting (see Figure 6.1).
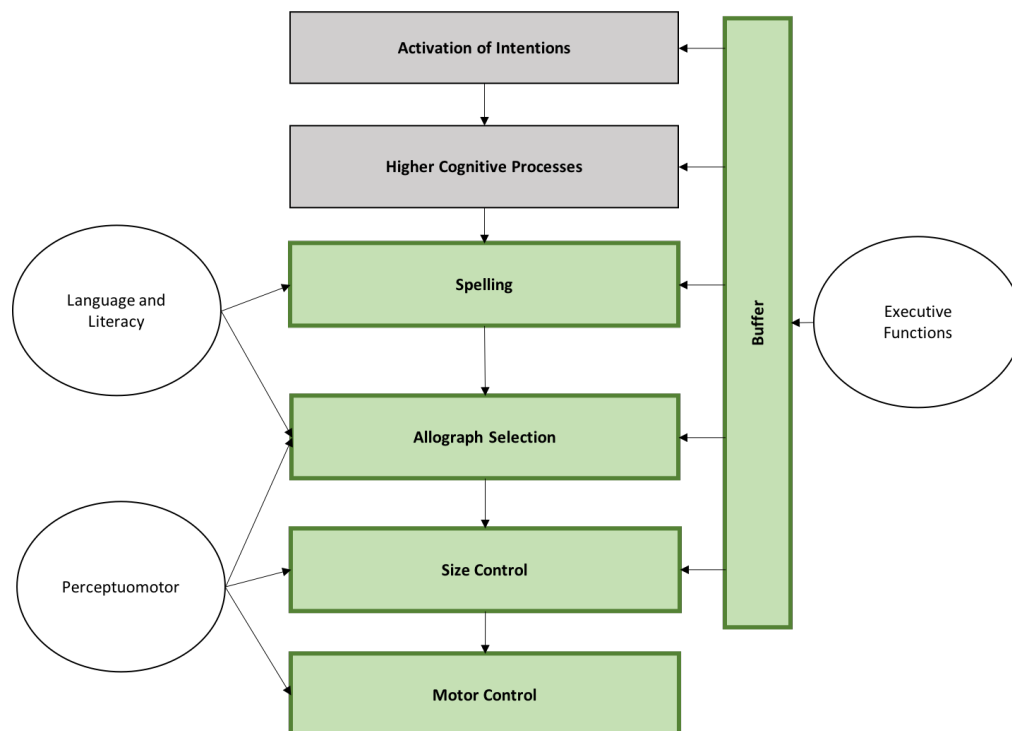
Figure 6.1. Modified psychomotor model of writing (van Galen, 1991) hypothesising how language and literacy, perceptuomotor, and executive function skills are related to handwriting production.

    6.1.1.1. Language and Literacy skills and handwriting. The psychomotor model (van Galen, 1991) outlined a simple set of processes to describe spelling. Spelling processes

has since been elaborated by a substantial body of work, which shows multiple linguistic components are important in spelling including, phonological, orthographic, morphological, letter, and lexical knowledge (Bahr et al., 2012; Bourassa & Treiman, 2003; Rapp et al., 2002). Also, since van Galen (1991) developed the psychomotor model of writing, converging evidence from experimental and individual difference studies has elaborated on how language and literacy influences handwriting production (Abbott & Berninger, 1993; Kandel & Perret, 2015).

Experimental studies investigating the effects of spelling complexity on handwriting have elucidated how spelling processes temporally influence handwriting fluency. In skilled adult writing, handwriting fluency has been found to be modulated by the lexical, orthographic, phonological, and morphological properties of the word being written (Delattre, Bonin, & Barry, 2006; Kandel et al., 2012; Roux et al., 2013). Collectively these findings show adults take longer to initiate writing, and are less fluent, when writing words with more complex spellings than less complex spellings. This means that spelling processes must be active during writing production and can render handwriting less fluent (Olive, 2014).

Spelling processes also influence handwriting fluency in children. Children as young as 8 years old took longer to produce strokes of letters (stroke durations) in words with a lower sound-to-spelling consistency than words with a higher sound-to-spelling consistency. Children also took longer to produce strokes of letters in lower frequency words than higher frequency words (Kandel and Perret, 2015). Thus, even in young writers, spelling processes appear to be active and exert some control over handwriting fluency.

In addition to the experimental studies of spelling processes, studies of individual differences report predictive associations between literacy-related skills and handwriting fluency (Abbott & Berninger, 1993; Berninger et al., 1994; Berninger et al., 1992). Abbott and Berninger (1993) examined the relationship between orthographic coding (letter, letter

cluster, word recall and homophone choice) and fine motor (finger succession, lifting, and spreading) skills on handwriting fluency (alphabet writing and sentence copying) amongst children in Grades 1 to 6 (equivalent to Years 2 to 7 in the UK). Interestingly, only orthographic coding skills significantly predicted handwriting fluency in all year groups and this association was stronger in older children (Grades 4 to 6/Years 5 to 7) than in the younger children. It is important to note that the absence of a direct relationship between fine motor skills and handwriting is somewhat surprising and is given further thought below. Nevertheless, the authors interpreted the predictive effects of orthographic skills on handwriting fluency as highlighting that language as well as motor skills contribute to handwriting fluency.

Few studies have examined the relationship between handwriting legibility and language and literacy abilities. Notably, though, using the SaHLT, we found letter formation legibility was related to literacy skills in children in Years 3, 4, and 5 suggesting that this aspect of legibility does tap literacy-related processes (see Study 1). Letter formation likely draws on multiple aspects of knowledge including letter's motor programmes as well as their phonological forms (Bara, Gentaz, Colé, & Sprenger-Charolles, 2004; Palmis et al., 2017; van Galen, 1991). The multimodal nature of letter formation is highlighted in Figure 6.1 by the arrow from language and literacy to allograph selection – in addition to perceptuomotor ability – because allograph selection is associated with letter formation (Prunty & Barnett, in press). Taken together, experimental and individual difference studies present converging evidence that handwriting is affected not only by how complex the spelling is but also by individual differences in children's language and literacy skills.

6.1.1.2. Perceptuomotor skills and handwriting. Following the activation of a spelling process, van Galen (1991) elaborates on three separate processing units which handle the more motor-based aspects of handwriting production (see Figure 6.1). The motor

programme (an abstract representation of an allograph's motor sequence) is retrieved from long term memory, and the size, speed, and spatial aspects of the letter are coded and implemented by the neuromuscular system (Graham, Struck, Santoro, & Berninger, 2006; Palmis et al., 2017; van Galen, 1991). Perceptual and motor skills contribute to these aspects of handwriting production. Since we found perceptual and motor skills to load onto a single dimension in Study 2, perceptual and motor skill are considered under the umbrella of perceptuomotor processes here.

Although precise estimates vary, at 7 years old children are believed to integrate perceptual (visual and kinaesthetic) information to provide online feedback during writing. Children rely heavily on perceptual feedback during writing prior to the stabilisation of motor programmes after the age of 11 years old but adults continue to use some perceptual feedback even after this stabilisation phase (Palmis et al., 2017; Thibon et al., 2018). Evidence demonstrating the importance of perceptual feedback for online correction during letter formation comes from studies of adults and children (see Danna & Velay, 2015 for review). For example, it has been observed that temporarily removing visual feedback leads to atypical handwriting behaviours such as longer and larger stroke trajectories in adults (van Doorn & Keuss, 1992). In children, temporarily removing visual feedback has resulted in increased pen pressure. This increase in pen pressure is assumed to reflect an increase in the amount of kinaesthetic feedback the child accessed to compensate for the loss of visual feedback (Chartrel & Vinter, 2006 as cited in Alamargot & Morin, 2015).

The ability to integrate visuospatial and motor information – when measured using a visual motor integration test – has been consistently associated with handwriting fluency and legibility (Cornhill & Case-Smith, 1996; Tseng & Chow, 2000; Tseng & Murray, Elizabeth, 1994; Volman, van Schendel, & Jongmans, 2006; Weintraub & Graham, 2000). Children with less legible and fluent handwriting tend to have poorer visual motor integration abilities

(Cornhill & Case-Smith, 1996; Tseng & Chow, 2000; Volman et al., 2006). Furthermore, visual motor integration ability explains a significant amount of variance in handwriting legibility and fluency in children with poorer handwriting. On the other hand, fine motor skills appear to be the most consistent predictor of handwriting legibility and fluency in children with better handwriting abilities (Tseng & Chow, 2000; Volman et al., 2006). Thus, visual motor integration is utilised in handwriting production and it appears to be particularly important in children with poorer handwriting, possibly because these children have not developed stable motor representations of letters (Palmis et al., 2017).

Following Berninger and colleagues (e.g., Abbott & Berninger, 1993) earlier work in predicting variation in handwriting fluency, Weintraub and Graham (2000) examined whether age, orthographic coding (alphabet writing, string matching, and letter, letter cluster, and word recall), finger function (succession, lifting, and recognition), and visual motor integration predicted the categorisation of children with good or poor legibility in fifth grade (equivalent to Year 6 in the UK). The authors found that both finger function and visual-motor integration – but not orthographic coding – predicted handwriting status, suggesting that visual motor integration and finger function are important skills for handwriting legibility. Taken together, the findings suggest visual motor integration ability is related to handwriting fluency and legibility in developing writers. In addition, the findings highlight that fine motor skills are also determinants of handwriting legibility and fluency.

The direct contribution of fine motor skills to handwriting ability contrasts with the earlier findings from Abbott and Berninger (1993) who found orthographic coding but not finger function directly predicted fluency. Weintraub and Graham (2000) used the same measures of orthographic coding and finger function as Abbott and Berninger (1993) yet reported the opposite findings. The contrast in findings could be due to methodological issues with the measures. Notably, the finger function tasks had poor correlations in the lower

grades (Abbott and Berninger, 1993; but see also Berninger & Rutberg, 1992). Alternatively, the divergence in correlates between handwriting legibility and fluency could suggest that the two handwriting skills tap different skills. Previous comparisons have found a weak relationship between handwriting legibility and fluency (Volman et al., 2006) and the two skills take on different developmental trajectories (Graham et al., 1998) highlighting the importance of assessing both fluency and legibility (see Chapter 1).

In addition to fine motor skills, other aspects of motor skill including posture, arm, and wrist control have also been reported to be important for handwriting production (Feder & Majnemer, 2007; Rosenblum, Goldstand, & Parush, 2006; Thibon et al., 2018). Children who had poorer posture were more likely to have poorer handwriting fluency (Rosenblum et al., 2006). In sum, visuospatial (visual motor integration) and fine motor skills in conjunction with postural stability and gross motor skills are related to handwriting production. Individual differences in these skills are associated with variances in handwriting legibility and/or fluency.

6.1.1.4. Executive functions and handwriting. Theoretically, the executive functions of memory and attention have been linked to writing. In the psychomotor model, van Galen (1991) posited that the outcome of each of the processes – including spelling and motor processes – was temporarily stored in separate aspects of working memory, which act as buffers between processes. Cognitive models of writing development also suggest memory and attention serve as processes necessary for directing writing, including handwriting. For example, Berninger and Winn (2006) considered working memory to be a central process for accessing long-term memory during planning, composing, reviewing, and revising writing. In addition, attention skills assist in the management of the writing process (Berninger & Antman, 2003; Berninger & Winn, 2006; Hayes & Berninger, 2014). Selective attention has been proposed as being utilised for specific aspects of the writing activity, whilst

ignoring/inhibiting other aspects. More recently, Hayes and Berninger (2014) described both memory and attention as resource level functions for writing to be called on by processes, including handwriting as part of transcription. Theoretically, it is clear that executive skills contribute to writing processes which includes handwriting. Empirical evidence also suggests the executive functions of memory and attention are related to handwriting production.

      6.1.1.4.1. Memory and handwriting. Empirical evidence supports theories that propose a link between memory and handwriting. Swanson and Berninger (1996) reported that verbal memory contributed a significant amount of variance in handwriting fluency amongst children in Grades 4 to 6 (Years 5 to 7 in the UK). The measures used to index verbal memory (nonword span, nonword written recall) tapped heavily on phonological and orthographic processes as well as memory (e.g., verbal/written nonword recall) which raises questions as to whether the variance in handwriting was explained by memory or by language skills (see earlier section). However, a more recent study by Kim and Schatschneider (2017) found verbal memory – indexed using a verbal span task with a lower phonological and orthographic component – explained a similar amount of variance in handwriting fluency amongst younger children Grade 1 to that reported by Swanson and Berninger (1996). Therefore, memory skills – regardless of whether they tap phonological orthographic skills – appear to be related to handwriting (fluency).

      6.1.1.4.2. Attention and handwriting. Relatively fewer studies have examined the relationship between attention and handwriting (Hooper et al., 2011). Studies with non-clinical samples have reported that children with less fluent handwriting perform more poorly on measures of sustained attention (Tseng & Chow, 2000). However, the strongest evidence for a link between attention and handwriting comes from neuropsychological studies describing handwriting difficulties in children with ADHD, a disorder characterised by impairments in attention. Children with ADHD without comorbid DCD performed worse

than typically developing children on several fluency and legibility measures when writing the same Hebrew letter repeatedly eight times. Children with ADHD produced letters that were larger and wider, more inconsistent in their height, and corrected more frequently than those of typically developing controls (Adi-Japha et al., 2007).

Similar findings have been replicated in children with ADHD when writing the letter < l > repeatedly. When writing the letter four times consecutively, children with ADHD without comorbid DCD produced stroke length that was more variable than that of typically developing children. Moreover, the variability in stroke length was strongly associated with ratings of inattention and total ADHD symptoms, suggesting handwriting difficulties were worse with increasing ADHD severity (Langmaid, Papadopoulos, Johnson, Phillips, & Rinehart, 2014). Therefore, impairments in attention abilities appear to be related to handwriting difficulties suggesting that attentional processes are implicated in handwriting production.

Due to the wealth of literature surrounding the role of memory in writing production and the relatively smaller focus on the role that attention plays, both memory and attention have been discussed separately. There is also some conflation between memory and attention in theoretical models of writing (see Chapter 1) and both are considered executive functions (see Study 2). Therefore, in this study, we consider memory and attention together as executive functions. It is clear than that executive functions contribute to writing processes and appear to be related to handwriting production.

The evidence reviewed here provides a theoretical framework with empirical evidence demonstrating how language and literacy, perceptuomotor, and executive functions skills influence handwriting production. Children with dyslexia, DCD, and comorbid dyslexia and DCD have deficits in one or more of these skills (Study 2) and have handwriting difficulties (Berninger et al., 2008; Blank et al., 2012). Indeed, some authors have suggested

a link between impairments in skills related to handwriting and handwriting fluency difficulties in children with dyslexia or DCD (Prunty et al., 2013; Sumner et al., 2013). In what follows, studies describing handwriting fluency and legibility difficulties in dyslexia and DCD and the relationships between handwriting difficulties and associated skills in these disorders are discussed.

6.1.2. Handwriting Fluency in Dyslexia and DCD

Handwriting fluency refers to the speed with which individuals produce written output. Using fluency measures as an index of handwriting ability has become more popular than using legibility measures. This is partly because fluency is easier to quantify and objectively measure than legibility (Abbott & Berninger, 1993; Graham, 1986; Rosenblum et al., 2003). Fluency is typically assessed using simple writing tasks under timed conditions where the main outcome measure of these tasks is to count the number of letters/words produced within a short time frame (Abbott & Berninger, 1993; Barnett et al., 2007). Advances in technology also allow online process measurements of fluency such as speed and pausing behaviours via pen tracking (see Chapter 1). The most popular task for assessing handwriting fluency is the alphabet writing task which involves children re-producing the letters of the alphabet in their correct sequence over a short time frame (Abbott & Berninger, 1993; Alamargot, Caporossi, Chesnet, & Ros, 2011; Alamargot & Morin, 2015; Berninger & Rutberg, 1992; Jones & Christensen, 1999; Pontart et al., 2013). Dysfluent handwriting as measured by these tasks has been reported in samples of dyslexia and DCD separately.

6.1.2.1. Dyslexia. Several studies describe handwriting fluency impairments amongst children with dyslexia (e.g., Berninger et al., 2008; Sumner et al., 2014; but see also Martlew, 1992) and report that difficulties persist into adulthood (Connelly et al., 2006). When writing the alphabet from memory, children with dyslexia wrote fewer legible letters than typically developing children (Berninger et al., 2008). The number of letters written in 15 seconds by

children with dyslexia was predicted by their selective attention/inhibition ability. Sumner et al. (2013) also administered an alphabet writing task and recorded performance over 60 seconds using a digitising tablet. Unlike Berninger et al. (2008), children with dyslexia in Sumner et al. (2013) wrote a similar number of letters in one minute to age-matched controls. Further analysis of the pen tracking data showed that despite writing a similar number of letters, children with dyslexia spent more time pausing than age-matched controls and this pausing was a similar duration to younger spelling-matched controls. When considered together, the findings from Sumner et al. (2013) help build a detailed picture of performance amongst children with dyslexia when writing the alphabet task. It suggests that children with dyslexia were able to form letters quickly, but their fluency was slowed by longer pauses, possibly because of delays in recalling the sequence of the alphabet.

Although not directly comparable, both Berninger et al. (2008) and Sumner et al. (2013) highlighted that children with dyslexia had writing fluency difficulties in what is considered an automatized task (Pontart et al., 2013). Fluency impairments early in this task amongst dyslexics appeared to be related to, in part, attentional (executive function) deficits, but fluency over a longer period appeared to be constrained by poorer alphabet sequence knowledge, a literacy related skill. The claim that literacy skills were constraining fluency in dyslexics was based only on the similarities in pausing between dyslexics and younger children, however, studies of handwriting fluency in word and sentence writing have shown more direct evidence for this argument.

When copying a short sentence from the DASH (Barnett et al., 2007) in their best handwriting (Copy Best) and again at speed (Copy Fast) children with dyslexia made longer pauses within words than typically developing age matched controls (Sumner et al., 2014). In children with dyslexia – but not typically developing controls – pausing duration within-words on both tasks was consistently highly correlated with reading and spelling ability. In

the Copy Fast task, pausing within words also produced large correlations with a composite measure of fine motor skill. However, the number of words written during the Copy Best task by children with dyslexia was uniquely predicted by spelling ability and pausing durations but not by fine motor abilities. The longer pausing durations– and therefore less fluent writing – of children with dyslexia was most strongly associated with spelling impairments; however, the mechanism by which spelling deficits result in longer durations is not clear.

Further experimental pen- and eye-tracking evidence provides greater specificity of how deficits in spelling processes impair handwriting within letters. Using a similar paradigm to that employed in their previous studies (Kandel & Perrett, 2015; Roux et al., 2013), Kandel et al. (2017) compared stroke durations rather than pausing durations between children with dyslexia and typically developing controls when copying words varying in consistency and frequency. Both children with dyslexia and typically developing children made longer stroke durations when copying inconsistent words than consistent words, but children with dyslexia made much longer durations than controls when the words were inconsistent. Furthermore, the fluency of children with dyslexia was specifically impaired by the lexicality of the word whereby dyslexics made longer movement durations when copying non-words. In addition to pen tracking, gaze data showed that children with dyslexia made more gaze lifts back to the model for low frequency, inconsistent words, and non-words. Together, these findings suggest that orthographic/phonological processing deficits in children with dyslexia result in decreased writing fluency.

Children with dyslexia have handwriting fluency difficulties at the alphabet, word, and sentence levels. These impairments could be partly related to deficits in executive function (Berninger et al., 2008), but are most strongly linked to deficits in literacy-related processes (Kandel et al., 2017; Sumner et al., 2014). It appears that spelling difficulties result in longer pauses within words when copying sentences (Sumner et al., 2014). It maybe that

longer pauses reported by Sumner et al. (2014) reflect more frequent look backs to the model because of orthographic/phonological processing deficits (Kandel et al., 2017). Orthographic/phonological processing deficits also result in decreased stroke fluency amongst children with dyslexia (Kandel et al., 2017).

Some theorists would argue that these findings are best explained according to a limited capacity account (see Chapter 1, p. 21). According to this view, impaired/unautomatized orthographic/phonological processing places a high cognitive cost on a limited pool of resources meaning fewer resources can be devoted to other writing processes operating in parallel such as graphomotor production (McCutcheon, 2011, Sumner et al., 2013). However, such findings could also be explained equally as well in the context of the interference account whereby delays in orthographic/phonological processing spreads into graphomotor production. Regardless of the action by which spelling processes act on handwriting production, handwriting fluency difficulties in dyslexia are most strongly related to literacy related processes.

6.1.2.2. DCD. Several studies have identified that children with DCD have marked difficulties in writing fluently (e.g., Rosenblum & Livneh-Zirinski, 2008; Prunty et al., 2013). Children with DCD tend to write fewer words per minute than typically developing children (Rosenblum & Livneh-Zirinski, 2008; Prunty et al., 2013). Similarly, to dyslexics, children with DCD also pause for longer when writing, reducing their global handwriting fluency on writing tasks (Prunty et al., 2013, 2014; Rosenblum & Livneh-Zirinski, 2008).

Assessing fluency amongst children with DCD using the alphabet writing task has produced inconsistent findings (Prunty & Barnett, 2017; Prunty et al., 2013; Rosenblum & Livneh-Zirinski, 2008). In some studies, children with DCD have been found to write fewer letters but have similar pausing durations to typically developing children (Prunty et al., 2013), others have found that children with DCD make longer pauses (Rosenblum & Livneh-

Zirinski, 2008), and others still fail to find a significant difference between children with DCD and controls on the number of letters written or time spent pausing (Prunty & Barnett, 2017). These inconsistent findings could be explained by the task itself. The alphabet writing task draws heavily on alphabet sequence knowledge (Pontart et al., 2013) which could be a relative advantage for children with DCD who do not have literacy related deficits (see Study 2); thus, strong alphabet knowledge could offset any fluency difficulties caused by deficits in motor skills. Indeed, handwriting fluency impairments have been described in children with DCD on tasks which do not require alphabet sequencing knowledge.

When completing the Copy Best and Copy Fast tasks from the DASH, children with DCD wrote fewer legible words per minute and spent a greater proportion of their time pausing than typically developing children (Prunty et al., 2013). Similarly, when copying two sentences, Rosenblum and Livneh-Zirinski (2008) reported that children with DCD spent significantly more time with their pen in the air (pausing) than controls. Thus, it appears that when children with DCD have fluency impairments, it is due to atypical pausing behaviour.

Parallels in pausing behaviour can be drawn between DCD in Prunty et al. (2013) and dyslexia in Sumner et al. (2013). In both studies, children with dyslexia and DCD spent more time pausing than typically developing children which suggests similarities in fluency difficulties between the disorders. On the basis of pause locations and correlations, Sumner et al. (2013) concluded that spelling-related deficits were responsible for dyslexics prolonged pauses however, Prunty et al. (2013) found no relationship between pausing durations and literacy measures in their DCD sample, instead suggesting that pausing was related to difficulties in automatizing handwriting due to their deficits in motor processes.

To further probe pausing behaviour in DCD, Prunty et al. (2014) examined performance on a composition task. Delineating processes involved in compositions is inherently more complex than simpler tasks such as copying because composition engages

higher level processes such as planning. When composing, children with DCD paused for longer within words than between words – in a similar manner to children with dyslexia – and spent a larger amount of time pausing within illegible words. Interestingly, when pauses were segmented, children with DCD did not differ in the amount of time they spent pausing in the lower ranges (e.g., between 30 – 250ms; believed to represent letter level processes) but they made more frequent very long pauses (> 10000 ms) than typically developing children. The frequency of these long pauses was predicted by fine motor skills – which were impaired in this group – and not by spelling ability (Prunty et al., 2014). Taken together, these findings suggest that the likely source of the prolonged pausing amongst children with DCD was due to motor difficulties. However, the mechanism underlying longer pausing among children with DCD is unclear.

Prunty et al. (2014) interpreted their findings from a limited capacity view (e.g., McCutcheon, 2011) and proposed atypical pausing behaviours in children with DCD represented difficulties in parallel activation of higher- and lower-level processes. The authors suggest that a lack of automaticity in handwriting, presumably due to deficits relating to fine motor skill development, results in handwriting processes placing a high cognitive load on limited resources leaving fewer or no resources available for higher-level processes. Alternatively, impaired retrieval of motor programmes or deficits in using feedback control may also explain this pausing behaviour (Yu & Chang, 2010).

The literature reviewed here highlights that children with dyslexia and DCD have handwriting fluency impairments. These impairments are similar in some respects. Most notably, longer pausing durations characterise handwriting in dyslexia and DCD. However, the position and the relationship of pauses with handwriting related skills suggests different putative bases for handwriting difficulties in dyslexia and DCD. Moreover, a further two explanations may explain the overlap in fluency difficulties between dyslexia and DCD. The

first is that participants with comorbid dyslexia and DCD were influencing performance in these samples (see Study 2). Although the dyslexic group performed no differently from controls on tasks of manual dexterity in Sumner et al. (2013), 30% of the sample in Prunty et al. (2013; 2014) had standard scores < 85 on tests of reading and spelling indicating that these children may have had comorbid dyslexia.

A second explanation is that shared deficits in executive functions between the disorders (Study 2) may also contribute to fluency impairments in dyslexia and DCD. Berninger et al. (2008) found executive functions to be related to alphabet writing performance in children with dyslexia yet few studies have examined the relationship between fluency and executive functions in dyslexia and DCD. It is therefore imperative to directly compare children with dyslexia, DCD, and comorbid dyslexia and DCD's handwriting fluency profiles and to examine the relationships between handwriting fluency in these groups with a large range of handwriting related skills necessary involved in handwriting production.

6.1.3. Handwriting Legibility in Dyslexia and DCD

Handwriting legibility describes the quality of the written production. Comparatively fewer studies have examined handwriting legibility, particularly in relation to dyslexia and DCD. Despite several legibility measures existing, they are plagued by a lack of objectivity, ease of use, and low reliability (Rosenblum et al., 2003). These legibility measures are broadly categorised as global or analytic measures. Global scales provide an overall impression of how readable handwriting is whereas analytic scales examine different dimensions of handwriting that contribute to the readability (see Chapter 1, p. 31). Handwriting legibility in dyslexia and DCD has predominantly been assessed using global scales.

6.1.3.1. Dyslexia. It is often reported anecdotally that individuals with dyslexia have less legible writing (Cooke, 2002; Berninger et al., 2008; Martlew, 1992). Despite several reports of legibility difficulties, to date, no study has objectively examined handwriting legibility in dyslexics. Instead, many qualitatively describe the nature of legibility issues. For example, in a case study, Cooke (2002) noted that a student with dyslexia had difficulty in correctly forming letters. Similarly, Martlew (1992) noted that letter formation among children with dyslexia was poor and like younger children's formation. Apart from letter formation and overall legibility, no investigation has examined handwriting legibility profiles amongst children with dyslexia or investigated why children with dyslexia have poorer handwriting legibility.

As discussed previously, few studies have examined the relationship between language and literacy and handwriting legibility in typically developing children and this is the case for children with dyslexia as well. However, a finding from Study 1 is relevant for considering how handwriting legibility impairments in dyslexia. Specifically, in the first study of this thesis we established a relationship between letter formation legibility and reading and spelling amongst typically developing children. Letter formation likely taps phonological or letter-sound knowledge which is frequently impaired in children with dyslexia (Bara et al., 2004; Longcamp, Velay, Berninger, & Richards, 2016; Thompson et al., 2015). Difficulties in letter formation stemming from impaired letter-sound knowledge might explain the locus of poor letter formation reported anecdotally (e.g., Martlew, 1992). Yet, the association between literacy impairments and legibility deficits in children with dyslexia has not, to our knowledge, ever been formally tested.

6.1.3.1. DCD. Given that handwriting impairments are one of the primary reasons for referral of children with DCD to occupational therapy (Miller, Missiuna, Macnab, Malloy-Miller, & Polatajko, 2001), it is unsurprising that more is known of the handwriting legibility

impairments in DCD than in dyslexia. Using a combined global and analytic scale applied to a piece of copied text, Rosenblum and Livneh-Zirinski (2008) compared handwriting in Hebrew between children with and without DCD. On this scale, children with DCD had poorer spatial arrangement of their writing, had poorer global legibility, and erased and wrote over more letters (see also Rosenblum et al., 2013). However, they did not differ from controls on the number of unrecognisable letters, a measure related to letter formation. In English, the legibility of children with DCD also differed from that of typically developing controls. Using another global legibility measure – the Handwriting Legibility Scale (HLS; Barnett et al. 2018) – applied to a short composition, children with DCD received lower ratings on all aspects of legibility (global legibility, effort required to read, layout, letter formation, and alterations). Therefore, the HHE and HLS suggest that children with DCD have diffuse handwriting legibility difficulties.

Whilst children with DCD have consistent handwriting legibility problems, it is not clear what such difficulties relate to. Despite deficits in perceptuomotor skills being a marker for DCD (Study 2) and associated with handwriting production (see earlier) surprisingly few studies have examined the relationship between perceptuomotor skills and handwriting in children with DCD. Prunty et al. (2016) examined the association between visuospatial skills with handwriting fluency and legibility ability in children with DCD. Although children with DCD performed significantly worse on the visuospatial tests, performance on these tests was not strongly associated with handwriting legibility or fluency difficulties. Therefore, this raises questions about the nature of handwriting legibility difficulties in children with DCD.

In comparison to handwriting fluency, fewer studies have sought to understand profiles of handwriting legibility in dyslexia and DCD. Perhaps one roadblock to examining handwriting legibility in these groups is the lack of reliable, sensitive, and valid measures of legibility (Rosenblum et al., 2003). By directly comparing profiles using the SaHLT

(Caravolas & Downing, in prep.) an analytic measure of handwriting legibility which permits the examination of different aspects of legibility we hope to investigate whether dissociable differences in legibility exist. Furthermore, examining the relationship between handwriting legibility and skills related to handwriting will provide a first step into elucidating the potential bases of handwriting difficulties in these groups.

6.1.4. The Current Study

This study was concerned with directly comparing handwriting fluency and legibility profiles between children with dyslexia and DCD to establish whether dissociable differences in handwriting exist between the disorders and typically developing children. A related aim was to examine the association between handwriting difficulties and the disorder-specific and shared marker impairments between dyslexia and DCD that were identified in Study 2.

To address these aims we compared children with dyslexia, DCD, comorbid dyslexia and DCD, and typically developing age-matched controls on alphabet writing (fluency) and on legibility ratings from responses to the SaHLT (Caravolas & Downing, in prep.). Alphabet writing was selected as it is a well-used index of handwriting fluency and performance has varied between dyslexia and DCD in separate studies previously (e.g., Prunty et al., 2013; Rosenblum & Livneh-Zirinski, 2008; Sumner et al., 2013). To gain a better understanding of performance on alphabet writing, and to clarify some of the inconsistent findings reported in alphabet writing tasks we go beyond the typically used measures to conduct analyses of the types of errors made. The SaHLT's Legibility measure was used because it is an analytic measure which permits the examination of aspects of legibility that tap literacy skills.

6.1.4.1. Dissociable profiles of handwriting difficulties. We expected that children with dyslexia and DCD would demonstrate dissociable handwriting profiles. On fluency, children with dyslexia (singular and comorbid) were expected to write fewer letters and to pause for longer when writing the alphabet whereas children with DCD-only were not. On

legibility, we expected that children with dyslexia would perform less well than controls on dimensions that tap literacy-related processes, which would include letter formation (see Study 1). Based on a slightly larger literature, we expected that children with DCD (singular and comorbid) would perform less well than controls on all dimensions of legibility, reflecting difficulties with the motoric aspects of legibility.

      6.1.4.2. Relationship between handwriting and related skills. Based on the reviewed evidence, we anticipated that handwriting legibility and fluency difficulties would be most strongly associated with the disorder-specific marker impairments whilst shared markers may also be associated with handwriting difficulties in dyslexia and DCD. Specifically, handwriting fluency and legibility difficulties in dyslexia should be associated with phonological and literacy abilities whereas the handwriting difficulties of children with DCD should be associated with perceptuomotor abilities. Executive functions should be consistently associated with handwriting difficulties to a lesser extent in dyslexia and DCD.

6.2. Methods

6.2.1. Participants

      This study used data from Phases 1 and 2 (see General Methods). Children were classified using the same marker criteria reported in Study 2. To balance the groups, typically developing children were matched as closely as possible to a child with dyslexia, DCD, or comorbid dyslexia and DCD on age (at Phases 1 and 2), nonverbal IQ (NVIQ), class year, gender, and handedness. There was some overlap whereby children with different disorders were matched to the same typically developing child (i.e., one child with dyslexia and another with DCD were matched with the same typically developing child). The demographics of each group are reported in Table 6.1. Despite being matched as closely as possible, children with comorbid dyslexia and DCD had significantly lower NVIQ than the control and dyslexic groups.

Table 6.1.

Demographics of Children with Dyslexia, DCD, and Comorbid Dyslexia and DCD Matched with Typically Developing Controls

	Group				*F*	η_ρ^2	Post-Hoc Comparisons
	DYS	DCD	DYS + DCD	TD			
n	28	25	17	39			
Age (Phase 1)	111.36 (10.33)	110.6 (10.48)	112.88 (12.44)	109.59 (10.86)	0.39	0.01	
NVIQ[a]	9.89 (3.71)	8.52 (2.63)	6.6 (2.59)	9.45 (2.73)	4.51**	0.12	TD = DYS > DYS+DCD
Literacy							
Word Spelling[b]	78.54 (9.34)	103.92 (15.07)	73.82 (8.56)	110.87 (13.74)	58.54***	0.63	TD = DCD > DYS = DYS+DCD
Word Reading[b]	82.46 (7.15)	104.31 (9.15)	82.06 (7.15)	100.80 (12.88)	40.49***	0.54	TD = DCD > DYS = DYS+DCD
Motor							
Visual Motor Integration[b]	90.64 (12.59)	80.84 (12.60)	74.53 (9.96)	93.68 (8.98)	15.49***	0.31	TD = DYS > DCD = DYS+DCD
Coordination[b]	90.29 (6.78)	75.2 (9.76)	78.06 (10.79)	92.74 (8.58)	26.81***	0.43	TD = DYS > DCD = DYS+DCD

Notes. Standard deviations are reported in parentheses. DYS = dyslexia-only, DCD = DCD-only, DYS+DCD = comorbid dyslexia and DCD, TD = typically developing.
[a]Scaled scores. [b]Standard scores.
p < .05. **p* < .01. ***p* < .001.

6.2.2. Design and Procedure

The aim of this study was to investigate profiles and correlates of handwriting legibility and fluency in children with dyslexia and/or DCD. To examine profiles of handwriting, we used handwriting legibility data collected during screening at Phase 1 (see measures below) along with alphabet writing data collected during Phase 2, on average seven months later. Children produced handwriting samples for legibility measurements during class screening (see Study 1) whereas children completed the alphabet writing task individually in a quiet area of their school or at a testing room at Bangor University.

6.2.3. Measures

6.2.3.1. Handwriting legibility. Handwriting legibility was rescored using an updated version of the SaHLT (Caravolas & Downing, in prep). This included the same four dimensions described in Study 1, Letter Formation, Letter Spacing, Word Spacing, and Line Alignment. The criteria of the dimensions were as in the original version (see Study 1) but they were applied using a 5-point scale from 1 (illegible) to 5 (excellent) to each sentence, instead of the original 7-point scale (see Figure 6.2). Scores for each dimension were derived by averaging the ratings over the number of sentences completed by the child. For example, a child whose letter formation was scored as 4 (good) on six sentences and 3 (OK) on a further four sentences would receive a score of 3.6 on the Letter Formation dimension. The sum of the four-dimension scores indexed total legibility. To illustrate, a child whose average scores were 3.6, 4.2, 4.4, 3.8 for Letter Formation, Letter Spacing, Word Spacing, and Line Alignment, respectively would receive a total legibility score of 16.

Handwriting legibility was scored separately to spelling by a trained research assistant who was blind to the child's status. Analysis has revealed this scoring system to be a valid and reliable measure of handwriting legibility. Specifically, there was a strong correlation ($r = -.54$, $p < .001$) between the SaHLT and the Handwriting Proficiency Screening

Questionnaire (HPSQ; Rosenblum, 2008), a teacher questionnaire of handwriting legibility

and fluency. The handwriting legibility scoring further demonstrates good inter-rater (r_{ICC} =

.83, 95% CI [.70, .91]) and test-retest (r_{ICC} = .76, 95% CI [.57, .87]) reliabilities.

Letter Formation	
Excellent	2. The boys and girls planted a tree.
Good	8. The magisian transformed scarfs into bunnies.
OK	6. The night fort a tuff battle.
Poor	7. some girls were riding ther bicicles
Illegible	7. Sane Sorro girls were riding unep by Skor

Letter Spacing	
Excellent	1. The cars were red.
Good	4. A fearless man killed the biggest monster.
OK	4. A Slebs man killed the biggest monster.
Poor	9. That sigh said smoking was a kcaale.
Illegible	9. Whrad is a rellyer to Sorwthe.

Word Spacing	
Excellent	6. The knight fought a tough battle
Good	2. The boys and girls planted a tree.
OK	3. He playfl appy is runig around the order.
Poor	10. the ordicence Used the cavo pre formena
Illegible	8. The musishon brasfomted skarv's into bunny's

Line Alignment	
Excellent	4. A fearless man killed the biggest monster.
Good	2. The boy's and girl's planted a tree
OK	2. the boys a hd girls planted a tree.
Poor	3. Her playful puppy in runing around the Garden
Illegible	8. The moish a Tash and Shafa in Sr and

Figure 6.2. Representative examples of responses receiving a score on the five points

between illegible to excellent for each dimension.

6.2.3.2. Handwriting fluency. We used a variant of the alphabet writing task from the DASH (Barnett et al., 2007). Children wrote the alphabet in lowercase letters in the correct sequence as quickly and as legibly as they could in 1 minute. They wrote on a piece of lined paper attached to the top of a digitiser to facilitate the recording of online graphomotor (speed and pausing durations) measures.

Both online and offline measures of fluency were taken. For offline measures, we followed the guidelines published in the DASH manual and counted the number of legible, correctly-sequenced, lowercase alphabet letters. In addition, various error types were coded. These included proportion correct (incorrectly formed letter based on the Letter Formation dimension of the SaHLT), insertions (a letter that was in an incorrect position in the sequence), deletions (a letter that was not been included in the sequence), and reversals (a letter which has had its orientation reversed, e.g., b → d).

For online measures, we extracted the speed and pausing duration of each letter from Eye and Pen 2.0 (Alamargot et al., 2006). Speed was defined as the distance travelled by the pen in cm per second (cm/s). Pausing durations were identified when the pen was inactive (Olive, 2010) for 30ms or longer. A low threshold of 30 ms was used because alphabet writing is assumed to be a highly automatised activity (2013) and previous studies have found differences between children with and without dyslexia or DCD populations using this threshold (e.g., Prunty et al., 2013; Sumner et al., 2013).

6.2.4. Apparatus

The administration and recording of pen tracking data on the Alphabet Writing task was undertaken using Eye and Pen 2.0 (Alamargot et al., 2006). The programme was run on a Dell Precision M4800 15-inch laptop connected to a medium Wacom Intuos Pro digitising tablet (sampling area: 224 x 140mm) sampling at 200Hz. The digitising tablet was positioned horizontally with lined A4 paper attached over the top of the pad to mimic the way children

would write in their exercise books. Children wrote onto the paper using a compatible

Wacom Inking pen which acts as a digitising stylus as well as leaving a visible ink trace on

the paper. Participants were briefed to hold and write with the 'special pen' as if it were a

normal ballpoint pen. All children had experience with writing using this set up from

previous orientation exercises they had completed using the same set-up.

6.2.5. Data Analysis

This study had two aims. The first was to examine the profiles of handwriting

legibility and fluency difficulties amongst children with dyslexia and DCD and the second

was to elucidate the relationship between handwriting legibility and fluency and

phonological, literacy, motor, and executive function ability. Group comparisons on

measures of handwriting legibility and fluency between children with dyslexia, DCD,

comorbid dyslexia and DCD, and typically developing controls were used to address aim one.

Group comparisons were followed by correlations to examine the relationships between

handwriting and phonological speed, literacy, motor, and executive function factor scores

estimates derived in Study 2.

6.3. Results

6.3.1. Profiles of Handwriting Fluency

Handwriting fluency was assessed using a speeded alphabet writing task completed

on a digitising tablet. As such, we compared group performance on product (accuracy and

error) and process fluency measures.

6.3.1.1. Product measures. Handwriting fluency has been traditionally estimated by

counting the number of legible correctly sequenced letters (e.g., Barnett et al., 2006). In

addition to calculating the number of correctly sequenced letters, we also measured the

proportion of correctly formed letters, insertions, deletion, and reversals.

6.3.1.1.1. Correctly sequenced letters. The mean number of correctly sequenced

letters and the respective standard scores are presented in Table 6.2. Groups differed

significantly in the number of correctly sequenced legible letters produced in 1 minute after

controlling for NVIQ, $F(3, 98) = 14.96$, $p < .001$, $\eta_\rho^2 = .31$. Children with dyslexia (single and

comorbid) wrote significantly fewer legible and correctly sequenced alphabet letters than

typically developing children ($ps \leq .001$). Children with dyslexia-only also wrote

significantly fewer legible and correctly sequenced letters than children with DCD ($p = .01$).

The number of legible and correctly sequenced letters failed to differ significantly between

comorbid dyslexia and DCD and DCD-only ($p = .131$), between typically developing

children and children with DCD-only ($p = .355$), and between children with singular or

comorbid dyslexia ($p > .999$).

Table 6.2.

Raw and Scaled Scores of the Number of Correctly Sequenced and Legible Alphabet Letters

Written

	Dyslexia	DCD	Dyslexia and DCD	Typically Developing
Raw	25.82 (9.11)	36.52 (13.18)	22.71 (8.92)	43.18 (12.42)
Scaled	7.11 (1.26)	8.96 (1.81)	6.53 (1.81)	9.82 (1.78)

Note. Standard deviations are reported in the parentheses. Scaled scores were derived from
the DASH norms (Barnett et al., 2007).

6.3.1.1.2. Errors. In addition to the number of correctly sequenced legible letters, we

also examined the types of errors made. We measured four types of errors which were:

proportion of correctly formed letters (according to the DASH criteria), the number of

insertions, deletions, and reversals. A one-way ANOVA (with Bonferroni corrections) was

used to compare groups on the proportion of correctly formed letters and Kruskal-Wallis

(with Dunn tests and Bonferroni corrections) were used to compare groups on the median

number of insertions, deletions, and reversal errors.

6.3.1.1.2.1. Proportion of correctly formed letters. The proportion of letter formation errors for each group are presented in Figure 6.3. Groups differed in the proportion of correctly formed letters, $F(3, 104) = 5.75$, $p = .001$, $\eta_\rho^2 = .14$. Pairwise comparisons with Bonferroni corrections revealed that children with DCD-only made significantly more errors than typically developing children ($p = .001$). There was also a marginally significant difference between children with dyslexia and typically developing children ($p = .06$) but all other comparisons failed to reach significance.
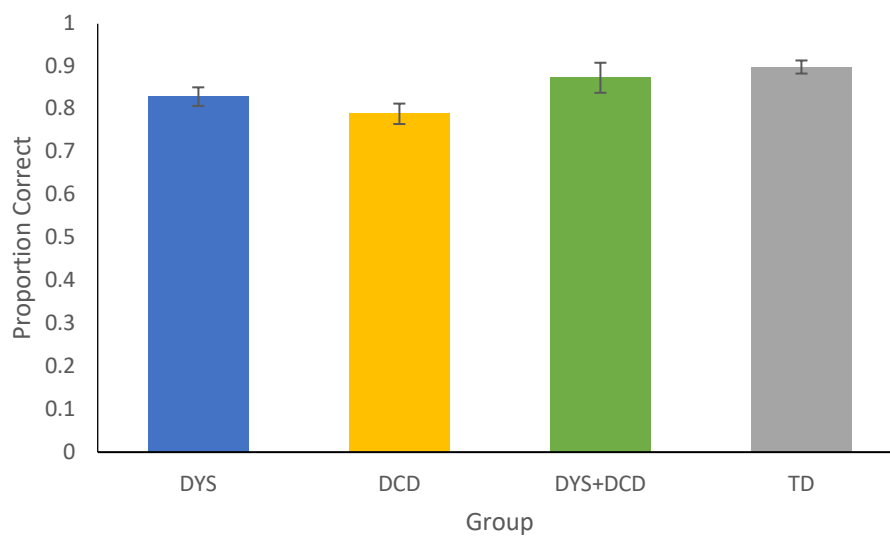
Figure 6.3. Proportion of correctly formed letters when writing the alphabet as a function of group. DYS = dyslexia, DCD = developmental coordination disorder, DYS+DCD = comorbid dyslexia and DCD, TD = typically developing. Error bars represent standard error.

6.3.1.1.2.2. Insertions. The median number of insertions, deletions, and reversals of each group are reported in Table 6.3. Groups differed significantly in the number of insertion errors made, $\chi^2(3) = 9.64$, $p = .019$. Children with dyslexia-only made significantly more insertion errors than typically developing children ($p = .031$). Children with comorbid dyslexia and DCD also made marginally more insertion errors than typically developing children ($p = .059$). Differences between typically developing children and children with

DCD failed to reach significance ($p > .999$) as did all other differences between the disorder

groups.

6.3.1.1.2.3. Deletions. Groups also differed in the number of letters that were not

included in responses (deletion errors), $\chi^2(3) = 26.71$, $p < .001$. Children with comorbid and

singular dyslexia made significantly more deletion errors than typically developing children

(ps < .05). Children with dyslexia (singular and comorbid) also made more errors than

children with DCD-only (ps < .05). Children with DCD and typically developing controls did

not differ in the number of deletion errors made ($p = .491$). There was no difference in the

number of errors made between children with singular and comorbid dyslexia ($p > .999$).

6.3.1.1.2.4. Reversals. All three disorder groups made a greater number of reversals

than typically developing children, however, comparisons of the median number of errors

failed to reach significance, $\chi^2(3) = 3.78$, $p = .286$.

Table 6.3.

Errors made by Children with Dyslexia, DCD, Comorbid Dyslexia and DCD, and Typically

Developing Children During Alphabet Writing

	Dyslexia		DCD		Dyslexia and DCD		Typically Developing	
	Mdn	95% CI	*Mdn*	95% CI	*Mdn*	95% CI	*Mdn*	95% CIs
Insertions	2	[0.81, 3.19]	0	[-1.04, 1.04]	1	[-0.95, 2.95]	0	[-0.72, 0.72]
Deletions	2.5	[-1.31, 6.31]	0	[-7.64, 7.64]	4	[0.99, 7.01]	0	[-0.95, 0.95]
Reversals	0	[-0.77, 0.77]	0	[-0.24, 0.24]	0	[-0.40, 0.40]	0	[-0.14, 0.14]

Note. 95% CI = 95% confidence interval.

6.3.1.2. Process measures. In addition to the offline measure, pen tracking was used

to record speed and pausing durations whilst writing each letter. Initial analysis of speed on

the whole response revealed extreme outliers for typically developing children and children

with comorbid dyslexia and DCD. These extreme outliers corresponded to illegible and

incorrectly sequenced letters and so the following analyses were undertaken on the speed and

pausing durations of correctly sequenced and legible letters only.

   6.3.1.2.1. Speed. The average correctly sequenced and legible letter writing speed per

group is reported in Figure 6.4. Writing speed was reduced in children with comorbid

dyslexia and DCD but speed did not differ significantly between groups, $F(3, 105) = 1.63$, p

$= .187$, $\eta_\rho^2 = .04$.
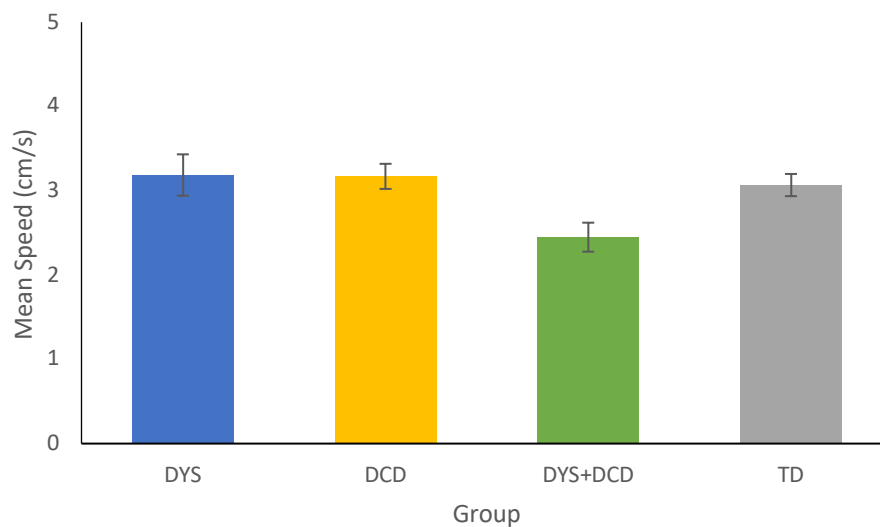
Figure 6.4. Mean speed when writing the alphabet as a function of group. DYS = dyslexia,

DCD = developmental coordination disorder, DYS+DCD = comorbid dyslexia and DCD, TD

= typically developing. Error bars represent standard error.

   6.3.1.2.2. Pause Durations. Average pausing durations of correctly formed and

sequenced letter for each group are reported in Figure 6.5. Analysis revealed a small but

statistically significant difference in pausing durations between groups, $F(3, 98) = 9.34$, $p <$

$.001$, $\eta_\rho^2 = .25$ after controlling for NVIQ ($F(1, 98) = 4.64$, $p = .034$, $\eta_\rho^2 = .05$). Children with

dyslexia made significantly longer pauses whilst writing than typically developing children (p

$= .001$) and children with DCD-only ($p = .001$). Children with comorbid dyslexia and DCD

also paused for significantly longer than children with DCD-only ($p = .044$). Pausing

durations did not differ significantly between controls and children with DCD ($p > .999$) or

between children with singular and comorbid dyslexia ($p > .999$).
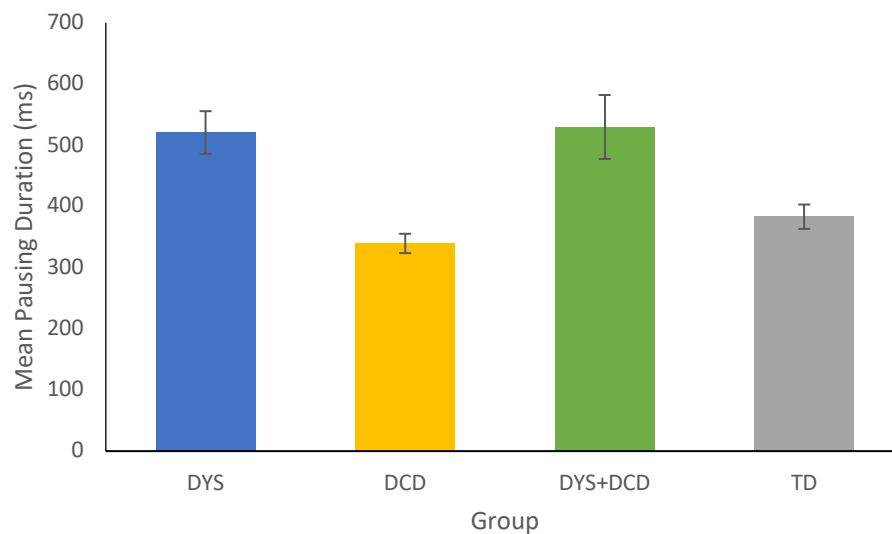
Figure 6.5. Mean pausing durations (ms) when writing the alphabet as a function of group.

DYS = dyslexia, DCD = developmental coordination disorder, DYS+DCD = comorbid

dyslexia and DCD, TD = typically developing. Error bars represent standard error.

6.3.2. Profiles of Handwriting Legibility

6.3.2.1. Total legibility. We scored children's responses to a sentence dictation task

on four dimensions of handwriting legibility using a 5-point scale. The total mean

handwriting legibility for each group is presented in Figure 6.6. There was a large significant

difference in the extent to which legibility differed between groups, $F(3, 104) = 22.94$, $p <$

$.001$, $\eta_\rho^2 = .4$. Children with dyslexia, DCD, and comorbid dyslexia and DCD received

significantly lower total legibility ratings than typically developing children ($ps < .01$).

Furthermore, children with DCD (singular and comorbid) received lower ratings than

children with dyslexia-only ($ps < .05$). Children with singular and comorbid DCD did not

differ significantly in their legibility scores ($p > .999$).
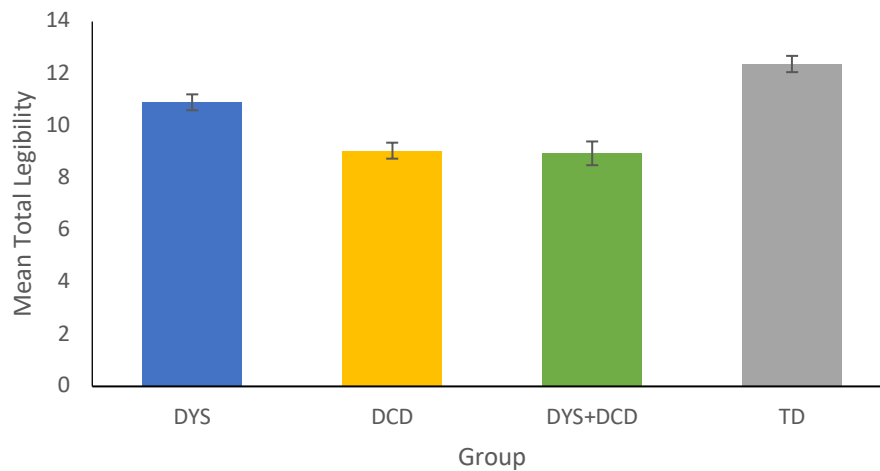
Figure 6.6. Mean total legibility scores as a function of group. DYS = dyslexia, DCD = developmental coordination disorder, DYS+DCD = comorbid dyslexia and DCD, TD = typically developing. Error bars represent standard error.

6.3.2.2. Individual legibility dimensions. Different dimensions of handwriting legibility are assumed to tap different handwriting processes. To better understand handwriting legibility in dyslexia and DCD we examined group profiles on the four handwriting legibility dimensions. A weighted MANCOVA was used to initially examine whether groups (IV) differed on the four dimensions (DVs). NVIQ was not a significant covariate in this or in subsequent analyses and so was removed from the models. Using Pillai's trace, groups differed across legibility dimensions, $V = .51$, $F(12, 309) = 5.23$, $p < .001$. Mean group legibility scores on each dimension are reported in Figure 6.7. We next examine group profiles for each dimension individually using oneway ANOVAs followed up with Bonferroni corrected pairwise comparisons. To compare the severity of handwriting impairments between disorder groups, we calculated the effect sizes (*d*) of significant pairwise comparisons.

6.3.2.2.1. Letter Formation. There were large differences between groups in letter formation legibility, $F(3, 104) = 19.00$, $p < .001$, $\eta_\rho^2 = .35$. Children with dyslexia-only

received much lower ratings for letter formation than typically developing children ($p < .001$, $d = 1.12$). Similarly, children with DCD also received lower ratings than controls ($p < .001$, $d = 1.54$) as did children with comorbid dyslexia and DCD ($p < .001$, 1.41). Ratings of letter formation legibility did not significantly differ between disorder groups (ps > .05).

6.3.2.2.2. Letter Spacing. Groups also differed on the letter spacing ratings, $F(3, 104) = 13.74$, $p < .001$, $\eta_p^2 = .28$. Children with DCD-only received significantly lower ratings for letters spacing than typically developing controls ($p < .001$, $d = 1.44$) and, to a lesser extent, children with dyslexia only ($p = .005$, $d = 0.97$). Similarly, children with comorbid dyslexia and DCD received lower letter spacing ratings than typically developing children ($p < .001$; $d = 1.13$) and children with dyslexia-only ($p = .052$, $d = 0.81$). Children with singular and comorbid DCD did not differ significantly, nor did children with dyslexia-only and typically developing children (ps > .05).

6.3.2.2.3. Word Spacing. Large group differences on word spacing ratings were also present, $F(3, 104) = 14.77$, $p < .001$, $\eta_p^2 = .30$. Children with DCD-only had much poorer word spacing ratings than typically developing children ($p < .001$, $d = 1.44$). Similarly, children with comorbid dyslexia and DCD achieved lower ratings than typically developing children ($p < .001$, $d = 1.39$). Interestingly, children with dyslexia-only also received moderately lower ratings than typically developing children on the word spacing dimension ($p = .05$, $d = 0.67$). Despite receiving a lower rating than typically developing children, children with dyslexia-only word spacing ratings were higher than children with DCD-only ($p = .043$, $d = 0.77$). Mean word spacing ratings of the comorbid group did not differ significantly from either dyslexia or DCD (ps > .09).

6.3.2.2.3.1. Word spacing post-hoc error analysis. There was no specific hypothesis regarding word spacing impairments and dyslexia. However, a potential explanation for impaired word spacing in children with dyslexia is that the word spacing dimension was

capturing *word boundary errors.* Word boundary errors describe instances where words are split into two plausible alternatives (hypersegmentation) or are combined (hyposegmentation), possibly at prosodic boundaries (Cutler & Butterfield, 1990). Such errors have been associated with reading, morphophonological, and phonological ability in young children (Correa & Dockrell, 2007; Tolchinsky, Liberman, & Alonso-Cortes Fradejas, 2015). In Slovak, children with dyslexia have been found to make the largest and most persistent spelling errors on words requiring boundary knowledge (Caravolas, Mikulajová, & Kuchaská, 2018). Therefore, it is possible that children with dyslexia's word spacing impairments were reflecting more frequent boundary errors. We tested this explanation by comparing the median number of words split into two plausible alternatives (hypersegmentations) and words combined (hyposegmentations) between groups using Kruskal-Wallis tests (χ^2).

Overall, the number of word boundary errors was small. On hyposegmentations, we found children with dyslexia (*Mdn* = 0), DCD (*Mdn* = 0), comorbid dyslexia and DCD (*Mdn* = 0), and typically developing children (*Mdn* = 0) did not differ significantly in the median number of words joined, $\chi^2(3) = 6.37$, $p = .095$. However, we did find that groups differed significantly in median frequencies of hypersegmentations, $\chi^2(3) = 18.22$, $p < .001$. Dunn tests with Bonferroni corrections revealed children with dyslexia-only (*Mdn* = 1) and children with comorbid dyslexia and DCD (*Mdn* = 1) split words into two plausible alternatives more frequently than typically developing children (*Mdn* = 0; *p*s < .001). Children with dyslexia-only also split marginally more words than children DCD-only (*Mdn* = 0; *p* = .057). There were no further significant comparisons. Hypersegmentation errors significantly correlated with phoneme blending in dyslexia ($r = -.39$, $p < .05$), but not in DCD ($r = .11$, $p > .05$), or typically developing children ($r = .04$, $p > .05$). Correlations between hypersegmentation

errors and reading measures did not reach statistical significance. Correlations were not run

with the comorbid dyslexia and DCD group due to the small sample size.

       6.3.2.2.4. Line Alignment. There was a smaller difference between groups on line

alignment legibility, $F(3, 104) = 11.85$, $p < .001$, $\eta_\rho^2 = .25$. Children with DCD-only had

poorer line alignment than typically developing children ($p < .001$, $d = 1.38$) and children

with dyslexia ($p = .006$, $d = 0.95$). Children with comorbid dyslexia and DCD also received

lower line alignment ratings than typically developing children ($p = .004$, $d = 0.96$) but

ratings did not differ significantly from children with dyslexia-only ($p = .182$). Children with

singular and comorbid DCD did not differ significantly on their ratings of line alignment, nor

did children with dyslexia-only and typically developing controls ($ps > .05$).
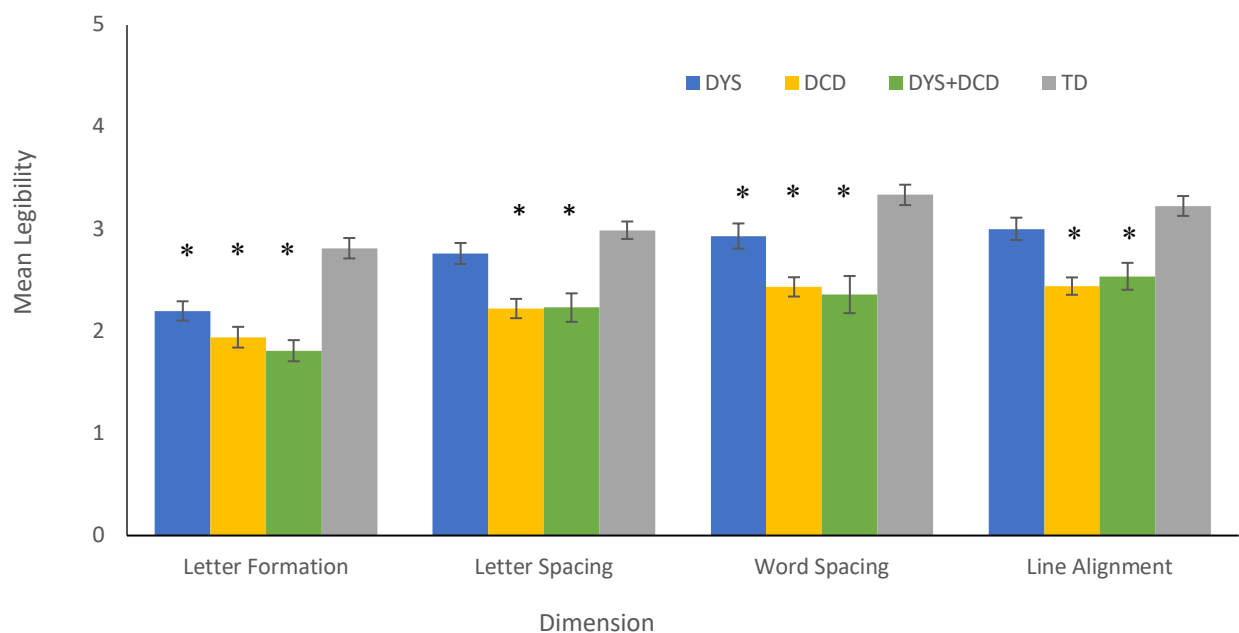
Figure 6.7. Mean ratings on each handwriting legibility dimension as a function of group.

DYS = dyslexia, DCD = developmental coordination disorder, DYS+DCD = comorbid

dyslexia and DCD, TD = typically developing. Error bars represent standard error.

*p ≤ .05

Analysis of group differences revealed separable profiles of handwriting difficulties in children with dyslexia and DCD. Children with dyslexia were less fluent and had poorer letter formation and word spacing, whilst children with DCD were not dysfluent, however, their legibility was poor on all dimensions. These profiles demonstrate a dissociation in the nature of handwriting difficulties which is consistent with the hypothesis that handwriting difficulties in dyslexia and DCD stem from different impairments. To further elucidate the nature of handwriting difficulties in dyslexia and DCD we examined correlates of handwriting fluency and legibility in these groups.

6.3.3. Relationships Between Handwriting Profiles and Related Skills

The nature of handwriting fluency and legibility impairments in dyslexia and DCD was probed further by examining the relationships between handwriting ability and skills related to handwriting in children with dyslexia and DCD. To do so, we ran Pearson's correlations between handwriting fluency and legibility measures and phonological, literacy, perceptuomotor, and executive function factor score estimates derived from the MIMIC model reported in Study 2. Patterns of correlations were examined separately for children with dyslexia-only, DCD-only, and typically developing children (see Tables 6.4 – 6.6 and Appendix C). Due to the small sample size of the group, children with comorbid dyslexia and DCD were not included in these analyses.

6.3.3.1. Dyslexia. Correlations between handwriting measures and handwriting related skills are reported in Table 6.4. The moderate positive correlation between total legibility and fluency suggests children with better legibility were also more fluent. There were also moderate phonological speed correlations with aspects of handwriting in which the dyslexic group showed impairments (Letter Formation, Word Spacing, and Fluency) and weaker (non-significant) associations with aspects of handwriting on which they were not impaired (Letter Spacing and Line Alignment). This pattern of correlations suggests that

handwriting impairments were related to deficits in phonological processing in children with dyslexia. A similar profile also emerged for the correlations between literacy and handwriting whereby literacy ability moderately-to-strongly correlated with aspects of handwriting that were impaired in this group (fluency, letter formation, and word spacing). However, there was also a strong correlation between literacy and line alignment, which was unimpaired in children with dyslexia.

There were also moderate-to-strong correlations between perceptuomotor scores – which were unimpaired in dyslexics – and all aspect handwriting legibility and fluency. Interestingly, the size of these correlations was larger than the associations between handwriting and phonological speed or literacy skills and likely reflect the strong motor component of handwriting. Finally, there were moderate correlations between executive skills and all handwriting fluency and legibility indices apart from letter spacing where the association was very weak.

Table 6.4.

Correlations between Handwriting Measures and Factor Score Estimates in Children with Dyslexia

	1	2	3	4	5	6	7	8	9
1. Letter Formation	-								
2. Letter Spacing	.42*	-							
3. Word Spacing	.33	.34	-						
4. Line Alignment	.57**	.46*	.54**	-					
5. Legibility	.74***	.74***	.76***	.78***	-				
6. Fluency	.41*	.25	.38*	.47*	.48*	-			
7. Phonological Speed	.31*	-.06	.42*	.23	.28	.50**	-		
8. Literacy	.51**	-.09	.38*	.53**	.42*	.49**	.57**	-	
9. Perceptuomotor	.67***	.39*	.49**	.58**	.69***	.49**	.36	.53**	-
10. Executive	.50**	.02	.48*	.48*	.47*	.59***	.92***	.65***	.66***

Note. Legibility = total handwriting legibility. Fluency = handwriting fluency indexed by the number of correctly sequenced legible letters in 1 minute.
***$p < .001$, **$p < .01$, *$p < .05$.

6.3.3.2. DCD. Correlations between handwriting measures and factor scores are reported in Table 6.5. Unlike for children with dyslexia, there was a weak and non-significant relationship between measures of impaired handwriting legibility and unimpaired fluency in this group. The dissociation in correlations between handwriting legibility and fluency also extended to the patterns of correlations between handwriting and handwriting related skills. That is, there were weak (non-significant) correlations between legibility – which was impaired in DCD – and phonological speed and literacy skills. Yet, handwriting fluency – which was unimpaired in this group produced strong correlations with phonological speed and literacy. In other words, unimpaired aspects of handwriting (fluency) were related to typical language and literacy ability in DCD.

The opposite pattern emerged in the correlations between handwriting measures and perceptuomotor skills, a DCD-specific marker (Study 2). Specifically, impaired handwriting

legibility – but not unimpaired handwriting fluency – strongly correlated with

perceptuomotor skills. Similarly, to children with dyslexia, handwriting fluency and legibility

moderately correlated with executive function.

Table 6.5.

Correlations between Handwriting Measures and Factor Score Estimates in Children with

DCD

	1	2	3	4	5	6	7	8	9
1. Letter Formation	-								
2. Letter Spacing	.51**	-							
3. Word Spacing	.44*	.61**	-						
4. Line Alignment	.68***	.55**	.59*	-					
5. Legibility	.81***	.80***	.79*	.87***	-				
6. Fluency	.22	.01	.18	.11	.14	-			
7. Phonological Speed	.30	.28	.34	.22	.35	.63***	-		
8. Literacy	.31	.25	.26	.26	.32	.79***	.90***	-	
9. Perceptuomotor	.51**	.66***	.57**	.56**	.64***	.18	.12	.09	-
10. Executive	.42*	.47*	.50*	.44*	.50**	.63***	.90***	.79***	.49*

Note. Legibility = total handwriting legibility. Fluency = handwriting fluency indexed by the
number of correctly sequenced legible letters in 1 minute.
***$p < .001$, **$p < .01$, *$p < .05$.

5.3.3.3. Typically developing. Table 6.6 shows there was a moderate correlation

between handwriting fluency and legibility. There were consistent moderate correlations

between phonological speed and all aspects of handwriting legibility and strong correlations

with fluency. Similarly, on literacy there were moderate-to-strong correlations with

handwriting legibility and strong correlations with handwriting fluency. In the main,

correlations between legibility and perceptuomotor and executive skills were weaker and

non-significant but the associations between fluency were moderate (perceptuomotor) or

strong (executive function).

Table 6.6.

Correlations between Handwriting Measures and Factor Score Estimates in Typically

Developing Children

	1	2	3	4	5	6	7	8	9
1. Letter Formation	-								
2. Letter Spacing	.65***	-							
3. Word Spacing	.45**	.56***	-						
4. Line Alignment	.52***	.63***	.54***	-					
5. Legibility	.80***	.86***	.78***	.82***	-				
6. Fluency	.37*	.33*	.32*	.19	.37*	-			
7. Phonological Speed	.37*	.35*	.31*	.36*	.36*	.73***	-		
8. Literacy	.50***	.43**	.34*	.33*	.45**	.59***	.87***	-	
9. Perceptuomotor	.24	.29	.10	.21	.28	.36*	.63***	.58***	-
10. Executive	.27	.28	.24	.15	.32*	.63***	.93***	.82***	.85***

Note. Legibility = total handwriting legibility. Fluency = handwriting fluency indexed by the
number of correctly sequenced legible letters in 1 minute.
***$p < .001$, **$p < .01$, *$p < .05$.

Comparisons of Pearson's correlation sizes between groups failed to reach statistical

significance in all cases which likely reflects a lack of power in the analyses. Therefore, we

interpret the difference in patterns of correlations across groups. The correlational analyses

revealed handwriting fluency and legibility were most strongly associated with phonological

and literacy abilities in typically developing children. However, different patterns of

correlations emerge in children with dyslexia-only and DCD-only. In dyslexia, only impaired

aspects of handwriting (fluency, letter formation and word spacing) were related to

phonological processing, suggesting handwriting impairments in dyslexia were related to

phonological processing deficits. A different pattern of correlations emerged amongst

children with DCD, where impaired aspects of handwriting were related to perceptuomotor

ability and unimpaired aspects of handwriting were related to phonological and literacy

ability. In both dyslexia and DCD, handwriting was related to executive function skills, but these correlations were not restricted to aspects of handwriting which were impaired.

It is important to emphasise that these analyses are correlational and so should be interpreted with caution. Given the current findings it would be prudent to conduct further analyses testing the relationships between handwriting fluency and legibility and related skills using regression or path analyses techniques. However, the relatively small sample sizes, particularly in the disorder groups, mean such analyses would have inadequate power (see Appendix D for power analysis of a simple path analysis on this data).

6.4. Discussion

This study was concerned with understanding handwriting difficulties frequently reported amongst those with dyslexia and those with DCD. We found dissociable profiles of handwriting difficulties and different patterns of associations with handwriting related skills between these disorder groups. The findings for fluency and legibility are discussed in relation to the relevant literature before turning to theoretical and practical implications and finally some limiting conditions.

6.4.1. Handwriting Fluency Impairments in Dyslexia and DCD

Alphabet writing is a popular task for indexing handwriting fluency and previous work had suggested that children with dyslexia and DCD perform differently on this measure. By examining performance on multiple aspects of this task we found dissociable differences between dyslexia and DCD. Using typically recommended scoring criteria (the number of correctly sequenced legible letters; Barnett et al., 2007) children with dyslexia were less fluent and paused for longer than typically developing children and children with DCD. These findings partially replicate other studies documenting prolonged pausing amongst children with dyslexia but near-typical performance amongst children with DCD (Sumner et al., 2013; Prunty & Barnett, 2017).

At first glance it would seem that children with dyslexia had fluency impairments on this task yet children with DCD did not. However, the common scoring criteria do not consider the number of total attempts made but does account for sequencing errors such as insertions and deletions. Analyses of these errors revealed children with DCD-only wrote fewer correctly formed letters out of their total number of errors but did not differ from typically developing children in the number of insertion or deletion errors. This suggest children with DCD did not have any difficulties in the sequencing component of the task. On the other hand, children with dyslexia made more letter insertions and deletions suggesting sequencing was a source of difficulty for these children.

Therefore, it is likely that children with dyslexia had poorer fluency on this task because they paused for longer when writing letters, possibly due to poor alphabet sequence knowledge. In contrast, children with DCD appeared as fluent as typically developing children despite having fewer legible letters. Children with DCD were able to achieve similar raw scores to typical developing children because their rapid (automatised) alphabet knowledge was able to offset the number of poorly formed letters. In this sense, strong alphabet knowledge was protective of fluency difficulties amongst children with DCD.

This interpretation is strengthened by the patterns of correlations between handwriting related skills and handwriting fluency. Fluency in children with dyslexia was strongly related to skills that are impaired in dyslexia – phonological speed and literacy, and executive functions – (Study 2) as has been reported previously (Berninger et al., 2008; Sumner et al., 2013) and suggests a link between disorder-specific and shared markers and handwriting fluency difficulties in this group. Interestingly, there was also a strong relationship between fluency and motor skills in the dyslexic group. A similar relationship in dyslexics has been noted previously (Sumner et al., 2014) and a weaker relationship was found also amongst typically developing children here. It is likely that this association reflected the motorically

demanding nature of the task. Yet, in children with DCD fluency was weakly associated with perceptuomotor skills, the latter being impaired in DCD, but was highly correlated with literacy skills adding weight to the thesis that alphabet knowledge was protective for children with DCD when completing this task.

When measured using the alphabet writing task, there is a dissociation in handwriting fluency between dyslexia and DCD. Performance on this task was most strongly associated with literacy skills, specifically alphabet sequencing knowledge. Children with dyslexia had fluency impairments on this task, possibly because of poorly automatized alphabet sequencing knowledge. Whereas the performance of children with DCD on this task was near typical – despite making a greater proportion of errors – possibly due to their stronger alphabet knowledge.

6.4.2. Handwriting Legibility Impairments in Dyslexia and DCD

Comparatively fewer studies have investigated handwriting legibility amongst children with dyslexia and DCD. To our knowledge, this was the first study to specifically investigate legibility difficulties in children with dyslexia. Notably, we also found a dissociation in profiles. Children with dyslexia had letter formation and word spacing impairments but typical letter spacing and line alignment whereas children with DCD (singular and comorbid) were impaired on all aspects of legibility.

It was anticipated that children with dyslexia would perform poorly on aspects of handwriting legibility that tap literacy related processes. As expected dyslexics had poorer letter formation because, in addition to the required motoric skills, this dimension of legibility is hypothesised to tap the fundamental literacy skill of letter-knowledge (Study 1). This is interpretation is consistent with the moderate to strong association between letter formation and phonological and literacy skills also observed in dyslexic children. An alternative explanation could be one of a coping strategy for poor spelling knowledge despite adequate

letter formation ability. That is, when writing unfamiliar words to dictation – as was done here – a child who is unsure of how to spell a word may consciously produce poorer letter forms to make it harder for the reader to discern whether the word has been spelt correctly or not.

In addition to letter formation impairments, children with dyslexia also had poorer word spacing. There was no specific hypothesis regarding word spacing as a direct link between this dimension of legibility and literacy skills has not been tested to date. Notably, the severity of the word spacing impairment in children with dyslexia was not as great as it was in children with DCD (singular and comorbid) suggesting word spacing difficulties in the dyslexic group were related to different impairments. We tested whether impaired word spacing in children with dyslexia was capturing *word boundary errors*. Indeed, we found children with dyslexia (singular and comorbid) more frequently split words into two plausible alternatives (hypersegmentations). The frequent hypersegmentations were related to phoneme awareness in this group. Previous findings have also highlighted that children with dyslexia have particular difficulties in word boundary knowledge (Caravolas et al., 2018) and that word boundary errors in young children are related to reading, morphological, and phonological abilities (Correa & Dockrall, 2007; Tolchinsky et al., 2014). Taken together, it is likely that word spacing impairments in dyslexia were the result of increased likelihood of splitting words inappropriately at prosodic boundaries reflecting poor morphophonological and possibly lexical knowledge. However, it is important to note that these were post-hoc in nature and further work is necessary to investigate these findings.

Unlike children with dyslexia, children with DCD were impaired on all dimensions of handwriting legibility, consistent with reports of diffuse handwriting legibility difficulties in DCD (Barnett et al., 2018; Miller et al., 2001; Rosenblum et al., 2013). Performance in this task was unrelated to language and literacy ability but was highly associated with impaired

perceptuomotor skills and – to a lesser degree – executive functions (Study 2). This finding is consistent with the notion that children with DCD have poorer handwriting due to their deficits in motor related skills but also acknowledges that deficits in executive functions may play a role.

Interestingly, there was a dissociation between legibility and fluency in children with DCD. The lack of association was evident in the direct correlation between these two aspects of handwriting and in the correlations between handwriting related skills and each of the two aspects of handwriting. This dissociation between legibility and fluency directly and in the relationships with related skills adds further evidence to task specific difficulties in this group. That is, children with DCD do not appear to have difficulties with tasks that require some motor input when they can use other (unaffected) handwriting related skills to meet the demands of the task. Similar findings have been reported amongst children with DCD in tasks beyond handwriting (Wilson et al., 2017).

6.4.3. The Nature of Handwriting Difficulties in Neurodevelopmental Disorders

The current findings contribute to our growing knowledge of the nature of handwriting difficulties in neurodevelopmental disorders affecting writing development. Specifically, the current investigation lends direct support to the thesis that handwriting difficulties are different in nature between dyslexia and DCD, a conclusion only ever implied previously. Furthermore, we find further (preliminary) evidence that handwriting difficulties reflect impairments in different underlying and associated skills in dyslexia and DCD. This strengthens the argument that the nature of handwriting difficulties is different in these disorders, and that deficits associated with each disorder are related to different expressions of handwriting difficulties. These findings also have important educational implications.

6.4.4. Implications for Assessment and Intervention

The current investigation has two important educational implications worthy of discussion. The first concerns the method for assessing handwriting fluency. The handwriting of children with DCD is often characterised as dysfluent (Blank et al., 2012); however, in the alphabet writing task, children with DCD seem to have compensated for motorically related fluency impairments with alphabet sequencing knowledge. This means that to gain a true estimate of handwriting fluency, examiners should use measures where handwriting fluency cannot be compensated by other skills.

A second educational implication relates to intervening with handwriting difficulties. The current study finds support for the notion that handwriting difficulties are related to deficits in different impairments in dyslexia and DCD. Therefore, any intervention that seeks to improve handwriting legibility and fluency should follow a complete assessment of the child's literacy, perceptuomotor, and executive skills. These assessments should then be used to identify areas of weakness to be targeted as part of the intervention. Thus, interventions will be individualised and should aim to improve handwriting related skills as well as handwriting directly.

6.4.5. Limiting Conditions

Prior to developing interventions, it is most important to fully establish causality. Notably, the current study examined the associations between handwriting and related skills via correlations and so causality cannot be inferred. Further work should examine the causal relationships between handwriting and related skills. In addition, further work should also consider the relationships between handwriting and related skills beyond correlations such as testing direct and indirect effects of related skills on handwriting. To do so, alternative analyses were considered, most notably path analysis. Whilst path analyses cannot establish

causality it can test direct and indirect relationships between handwriting and related skills, however, Monte-Carlo simulations revealed that power would be too low (see Appendix D).

In grounding the relationship between handwriting related skills and handwriting in van Galen's (1991) model, a unidirectional relationship between the handwriting related skills and handwriting has been inferred. Specifically, that spelling processes affect motor processes but motor processes do not affect spelling processes. Yet, the relationship between spelling and motor processes in developing children is likely to be bidirectional. A bidirectional relationship is evident in young children when training motor aspects of letter formation assists in developing letter knowledge (Bara et al., 2004; Bara et al., 2016; Longcamp et al., 2005) an important predictor of later literacy development (Caravolas et al., 2012).

6.4.6. Conclusion

This investigation was concerned with understanding profiles of handwriting fluency and legibility between children with dyslexia and DCD and the associations of these profiles with handwriting related skills. While there was some overlap, we established that there were different handwriting profiles of fluency and legibility impairments between children with dyslexia and DCD. The nature of handwriting impairments and their patterns of association with handwriting related skills suggested handwriting impairments were most likely to be associated with disorder-specific markers. Thus, the present findings suggest that handwriting difficulties differ according to disorder (and to task) and they reflect distinct marker impairments associated with the disorders.

Chapter 7

Study 4: Letter Formation Difficulties in Dyslexia and DCD: An Impairment in

Learning Orthographic Characters?

7.1. Introduction

Findings from Study 3 suggest separable profiles of handwriting legibility impairments between dyslexia and DCD. Interestingly, though, the dimension of *letter formation* legibility was judged to be the poorest in both dyslexia and DCD. This likely reflects the complexity of learning to form legible letters fluently. Learning to form letters takes several years to master (Thibon et al., 2018) and children must learn letter-specific visual, phonological, and motor knowledge. That is, children must build multi-modal representations of letters (Rothlein & Rapp, 2014). An impairment in acquiring knowledge in one or more of the modalities could explain, in part, the letter formation difficulties amongst children with dyslexia and/or DCD. In particular, developing suitable motor programmes of letters is a critical aspect of skilled handwriting production (Portier, van Galen, & Meulenbroek, 1990; van Galen, 1991). It is therefore important to investigate whether children with dyslexia and/or DCD have impairments in learning letter motor programmes, which could contribute to later letter formation difficulties.

7.1.1. Learning to Form Letters

The general motor learning framework is often used to describe handwriting development from a motor learning perspective (e.g., Palmis et al., 2017). Although many proponents of this framework discuss it in terms of general handwriting acquisition (e.g., Thibon et al., 2018), it actually describes more specifically how allographs or specific variants of letters (e.g., <l>, < L >, < ι > are all allographs of the same letter) are learned. That is, the framework is more concerned with how specific variants of letters are learned. This framework has already been evaluated in detail previously (see Chapter 1). For clarity, the motor learning framework of handwriting development is briefly recapped here.

The framework describes learning to form letters in terms of transitions between strategies beginning at the onset of formal instruction. Early in handwriting development, children use ballistic movements – which describes movements that reach maximum velocity quickly – to form strokes that comprise the letters. Ballistic movements limit the availability of feedback until the movement has finished. This means errors cannot be corrected until the end of the movement resulting in poor formation accuracy. Between the ages of 7 and 8 years old, children begin to integrate visual and kinaesthetic feedback which allows for online correction during letter formation. Finally, with experience in forming letters, at around 11 years old, dedicated motor programmes consolidate. As motor programmes consolidate and stabilise, children rely less on feedback and generate movements based on these motor programmes (Palmis et al., 2017; Thibon et al., 2018). Thus, to be able to form letters accurately and fluently, children must learn the properties of letters and develop an internal representation, or motor programme, of each allograph (Portier et al., 1990; van Galen & Teulings, 1983). Letters vary in several ways including their frequency of use as well as their visuomotoric properties, all of which affect the ease of acquiring and producing letters (Gosse, Carbonnelle, de Vleeschouwer, & Van Reybroeck, 2018).

7.1.1.1. Properties of letters. Allographs are not an arbitrary set of shapes but share similarities in their properties (Treiman & Kessler, 2011; Treiman, Levin, & Kessler, 2012). For example, we use similar curved and straight strokes to form the letters < p >, < q >, < d >, and < b >. Properties of letters influence the ease with which we acquire and produce letters. The visuomotor properties of letters include their orientation and motor complexity.

7.1.1.1.1. Orientation. A salient visual property of letters is the direction in which they face, or, more simply, their orientation. In the English alphabet, most letters comprise a

combination of a vertical stroke (*hasta*) accompanied by a body (*coda*) located on either the left

or the right (Brekle, 1994 as reported in Treiman & Kessler, 2011). It is the location of the coda

in relation to the hasta that provides letter's orientation. To illustrate, the letter < b > comprises a

coda located to the right of the hasta. Whereas, the letter < d > is constructed with the coda

located on the left of the hasta. For ease and following the convention set by Treiman and

Kessler (2011), in this study letters with a right hasta-coda orientation (e.g., < b >) are referred to

as *b-type* letters whilst those with a left hasta-coda type orientation (e.g., < d >) are referred to as

d-type letters. Of course, some letters do not have a clear orientation (e.g., < o >) and so are

referred to as *n*-type letters.

Young writers frequently reverse the proper orientation of letters to produce reversal

errors, sometimes referred to as mirror writing (Fischer, 2011; Treiman et al., 2014). Reversal

errors are typical in young children and the rates of errors rapidly decrease after the age of 6

(Fischer, 2011). Such errors have long been attributed to individual differences in children (e.g.,

Collette, 1979; Sala & Cubelli, 2007) although more recent evidence suggest reversal errors

reflect statistical irregularities in the letter's orientation (Fischer, 2013; Treiman & Kessler,

2011).

English alphabet letters predominantly have a b-type or d-type orientation. In fact, the

balance between the orientations of the letters is unequal with the majority of letters facing to the

right (a b-type orientation). Treiman and Kessler (2011) tested whether the orientation of the

letter influenced young children's (4 to 7-year olds) letter writing accuracy. They found children

made a greater number of errors and more often reversed letters with the less frequent, d-type,

orientation. This pattern has been replicated for letters as well as digits (Fischer, 2009; 2013;

Treiman et al., 2014).

The propensity for young writers to reverse lower frequency letters can be explained in the context of integrating multiple patterns (IMP) theory (Treiman & Kessler, 2014). According to this theory, children implicitly learn through exposure that letters are more likely to have a b-type orientation than a d-type orientation (see Chapter 1). That is, b-type letters have a higher probabilistic rate of occurrence. In the absence of certainty, young children rely on this information and form letters to be right-facing (Treiman & Kessler, 2011). Thus, at the beginning of instruction in writing, children are sensitive to the visual orientation of letters when forming them. They are more likely to form letters correctly if they have a higher probability b-type orientation. Children's sensitivity to the visual orientation of letters begins to wane with increasing age (Fischer, 2013). This decreasing sensitivity to the visual orientation of characters with age could be explained by the gradual acquisition of letter's motor programmes or even phonological information (Brooks, Berninger, & Abbott, 2011).

An important methodological note is that research on visual orientation in letter formation has examined performance using accuracy or error data. As accuracy improves with age, the sensitivity of accuracy/error analyses decreases. A more sensitive method for examining the effect of the letter's visual orientation on letter formation, particularly in older children, is to examine online fluency-based measures (e.g., speed, pausing etc.; see Chapter 1). Nevertheless, younger and less proficient writers are likely to be more strongly influenced by the orientation of the letters.

7.1.1.1.2. Motor complexity. The motor complexity of letters can be indexed in several ways (Meulenbroek & van Galen, 1990). A method for measuring motor complexity is to consider the number of strokes required to form a letter. That is, letters with a larger number of strokes are more complex than letters with fewer strokes. Similarly to orientation, proficient

(adult) writers are not influenced by the number of strokes in characters but young children are (Treiman & Yin, 2011; van Mier & Hulstijn, 1993).

Converging evidence suggests that young children are sensitive to the number of strokes within letters. For example, young Chinese writers accurately produced more characters with fewer strokes than those with more strokes. Children's name characters were more likely to be accurately recognised by adults if they had fewer strokes (Treiman & Yin, 2013). In addition to being less accurate when copying letters with more strokes, children's fluency – measured by the number of absolute velocity peaks where a fluent trace gives fewer velocity peaks – decreased as the number of strokes in letters increased (Thibon et al., 2018). Interestingly, increasing the number of strokes had a larger effect in decreasing fluency amongst 6- and 7-year-olds than 8- and 9- years old. Between 8- and 9-years of age, fluency plateaued, which the authors interpreted as a switch from stroke-by-stroke coding to coding larger units such as whole letters (Thibon et al., 2018).

Switching from stroke-by-stroke to larger coding units coincides with the shift from ballistic to perceptual feedback strategies at around the age of 8 years old (Maldarelli, Kahrs, Hunt, & Lockman, 2015; Palmis et al., 2017). It is also consistent with findings from skilled (adult) writers who code whole letters rather than on a stroke-by-stroke basis (Teulings et al., 1983; van Mier et al. 1993). Thus, young writers are sensitive to the motoric complexity of the letters and this sensitivity appears to fade as children's motor programmes for letters begin to consolidate.

7.1.1.2. Visuomotor and phonological considerations. The evidence reviewed thus far suggests children are sensitive to the visual orientation and motor complexity (visuomotor) properties of letters. However, children do not learn the visuomotor properties of letters in

isolation, but they also learn the letter's names and sounds as well. That is, they acquire

phonological and graphemic knowledge of letters. It is therefore hard to discern whether children

are sensitive to the visuomotor and/or phonographemic properties of the letters. For example, in

some of the studies reported in Treiman and Kessler (2011), children were asked to write letters

to dictation. Children's ability to do this, even at a young age, suggests they had acquired some

phonological knowledge of the allograph. Some graphemes such < b > and < d > rhyme and so

reversals between these letters could reflect difficulties in integrating phonological and

orthographic knowledge rather than a sensitivity to the visual aspects of the letter (Brooks et al.,

2011). It is therefore important to control for phonological confounds when examining the

effects of visuomotor properties of letters on learning to form letters.

To recap, to form letters legibly and fluently, children must acquire visual, phonological,

and motor knowledge of the letters. Acquiring motor knowledge of letters involves learning a

motor programme for each allograph, which appears to be influenced by the visuomotor

properties of the character. The influence of these properties on production seems to decrease

with increasing experience in forming the letter, perhaps reflecting increasing consolidation of

the letter's motor programme. Letter formation difficulties in dyslexia and DCD (see Study 3)

could reflect impairments in learning motor programmes.

7.1.2. Letter Formation Difficulties in Dyslexia and DCD

In Study 3, both children with dyslexia and children with DCD had poor letter formation

legibility. This chimes with previous findings that letter formation is impaired in both dyslexia

and DCD (e.g., Berninger et al., 2008; Martlew, 1992; Prunty et al., 2013; Rosenblum & Livneh-

Zirinski, 2008; Sumner et al., 2014). However, these studies have considered letter formation in

the context of general handwriting ability and not focused on the specific nature of letter

formation impairments. As such, few studies have considered whether letter formation errors could reflect impairments in learning motor programmes of letters.

Impairments in learning motor programmes could render children with dyslexia and/or DCD more sensitive to the visuomotor properties of allographs in a similar manner to younger children. To our knowledge, only one study has examined learning letter motor programmes, and this was amongst children with DCD-only (Huau, Velay, & Jover, 2015). Furthermore, no study has examined whether children with dyslexia and/or DCD may be more sensitive to the visuomotor properties of letters whilst learning them. Some studies have reported that children with dyslexia and DCD make errors consistent with confusions of letter orientation and motor complexity which could tangibly support the suggestion that children remain sensitive to the visuomotor complexity of letters in a similar manner to younger children.

7.1.2.1. Visuomotor properties of letters and formation in dyslexia. In a similar manner to young writers, it is often reported that children with dyslexia make frequent reversal errors (Brooks et al., 2011; Fischer, Liberman, & Shankweiler, 1978). In these studies, the overall number of reversal errors was low, but children with dyslexia were more likely to make reversal errors (Brooks et al., 2011). More recent studies using large samples of typically developing children have found that children who, at the age of 5 years old, were poor spellers were more likely to make reversal errors than children who were not poor spellers (Treiman, Kessler, Pollo, Byrne, & Olson, 2016). Moreover, reversal errors made by children aged 5 or 6 years old explained a small but significant amount of variance in later spelling ability (Treiman, Kessler, & Caravolas, 2018; Treiman et al., 2016). This higher frequency of reversal errors and its relationship with spelling ability in young children suggests children with dyslexia may be more sensitive to the orientation of the letters.

However, the aforementioned studies do not elaborate on the direction of the reversal errors making it hard to establish whether reversal errors among poor spellers/children with dyslexia were motivated by sensitivity to frequency information about the orientation of letters. Another study by Treiman et al. (2014) examined the direction of reversals amongst 5-year-old children with and without speech sound disorder (SSD). Children with SSD are at increased risk of dyslexia when literacy instruction begins (Pennington & Bishop, 2009). They found that although children with SSD produced fewer legible letters than typically developing children, the groups did not differ on the frequency with which they made b-type or d-type reversal errors. Interestingly, reversal errors did not explain a significant amount of variance in later reading ability in this study either.

The findings by Treiman et al. (2014) contrast with the aforementioned studies (e.g., Brooks et al., 2011; Treiman et al., 2016) by finding no evidence of a relationship between reversal errors and literacy ability. Moreover, Treiman et al.'s (2014) findings also suggest children with dyslexia may not be more sensitive to the visual orientation of letters than typically developing children. A possible explanation for this discrepancy is that the increased sensitivity to the visual orientation of letters by children with dyslexia only becomes apparent when typically developing children begin to consolidate motor programmes. Given the young age of children in the study of Treiman et al. (2014), it is possible that the typically developing children had not begun to consolidated motor programmes sufficiently for detectable group differences. Notably, children with dyslexia in Brooks et al. (2011) – where more children with dyslexia reversed letters – were on average 7 years older. Recall that children only begin to fully consolidate motor programmes of letters after extensive practice (e.g., Palmis et al., 2017). Therefore, it could be that the sample in Treiman et al. (2014) was too young for group

differences to arise. Moreover, the potential influence of the motor complexity on letters learning amongst children with dyslexia remains unstudied.

7.1.2.2. Visuomotor properties of letters and formation in DCD. Surprisingly few studies have investigated whether children with DCD make letter formation errors based on the visual orientation of motor complexity of letters. However, a recent analysis by Prunty and Barnett (in press) examined in detail the nature of letter formation errors made by 10-year-old children with DCD on Alphabet Writing and Sentence Copying tasks from the DASH.

Prunty and Barnett (in press) reported children with DCD made a higher frequency of letter reversals than typically developing children (see also Rosenblum et al., 2013). This higher frequency of reversal errors could indicate that children with DCD had poorly consolidated motor programmes and thus remained sensitive to the orientation statistics of the letters. Unfortunately, the authors did not elaborate on whether children with DCD made more reversal errors on b-type or d-type letters making it hard to determine whether older children with DCD were still sensitive to the statistical variations of orientation.

In addition to the frequent reversal errors, Prunty and Barnett (in press) also found children with DCD were significantly more likely to miss strokes. Children with DCD were also more likely to make different types of formation errors (e.g., incorrect start position, additional strokes, and deleting strokes) when forming the same letter in different tasks. This increased frequency of missing letters and lack of consistency in forming the same letters between tasks is indicative of not yet possessing the proper knowledge to construct letters, possibly due to a problem in learning motor programmes.

Impairments in learning motor programmes of letter-like characters have been reported in children with DCD (Huau et al., 2015). In the study of Huau et al. (2015), children with DCD

and matched controls learned a new character by copying it six times using a visual reference and a further six times without a reference. During training, children with DCD were less accurate than controls and neither group's accuracy improved over learning trials. Both groups' fluency – measured by the number of stops, where fewer stops would render formation more fluent – increased during training; but groups did not differ in dysfluency. Although children with DCD were as fluent as controls, velocity profiles were more variable within this group suggesting difficulties in consolidating motor programmes.

It is difficult to relate the conclusions of Huau et al. (2015) to children's learning of English alphabet letters because the novel character in that study – with a d-type orientation and at least four strokes – was more complex than most alphabet letters. Moreover, the use of only one novel character makes it impossible to infer whether children with DCD were sensitive to visuomotor properties of characters. However, like Prunty and Barnett (in press), Huau et al. (2015) reported that children with DCD were less consistent when forming letters, suggesting difficulties in learning motor programmes.

7.1.3. The Current Study

In this study we investigated whether letter formation difficulties in dyslexia and DCD could stem, in part, from impairments in learning letters' motor programmes by probing learning patterns and rates of novel orthographic characters that vary in their visuomotor complexity. Impairments in learning motor programmes was addressed in two questions. The first, was whether children with dyslexia and/or DCD had impairments in learning novel characters. Learning in this study was primarily indexed using pausing durations – a proxy measure of handwriting fluency – whereby a reduction in pausing durations would suggest increased fluency and increased learning of the characters respective motor programme. Impairments in learning

motor programmes in dyslexia and/or DCD would be indicated by children making longer pause durations than typically developing children at follow up.

When learning a letter's motor programme, children appear to be sensitive to its visuomotor properties, with more visuomotorically complex letters being harder to learn. This sensitivity to the visuomotor properties of letters fades as children consolidate knowledge in the form of motor programmes. Therefore, the second question addressed whether children with dyslexia and/or DCD remained sensitive to the visuomotor properties of the characters later than typically developing children when learning the novel characters. It was expected that children with dyslexia and/or DCD would pause for shorter durations for visuomotorically less complex (e.g., b-type orientation with fewer strokes) after training, but typically developing children would not. Such a finding would indicate children with dyslexia and/or DCD remained sensitive to the visuomotor properties of the characters and further suggest impaired learning of motor programmes.

7.2. Method

7.2.1. Participants

This study took place during Phase 3 of the project (see General Methods). The same criteria were used to identify children with dyslexia-only, DCD-only, and comorbid dyslexia and DCD. To better balance the groups, typically developing children were matched as closely as possible to children with dyslexia, DCD, and comorbid dyslexia and DCD on age, non-verbal IQ (NVIQ), class year, gender, and handedness. In some cases, two or three children from the disorder groups were matched with a typically developing child. In total, data from 99 children were used this study (children with dyslexia: $n = 27$, children with DCD: $n = 23$, children with

comorbid dyslexia and DCD: $n = 15$, typically developing children: $n = 34$). The demographics

of each group are reported in Table 7.1.

Table 7.1.

Demographics of Children with Dyslexia, DCD, Dyslexia and DCD Matched with Typically
Developing Children Controls

		Disorder Group	Matched Controls	*t*
		Dyslexia		
n		27	24	
Age (months)		118.44 (10.44)	118.75 (9.69)	0.11
Non-verbal IQ[a]		9.89 (3.71)	9.58 (3.03)	0.32
Literacy[b]				
	Word Spelling	79.00 (9.18)	113.00 (13.69)	10.52***
	Word Reading	82.59 (8.53)	107.04 (9.38)	9.74***
Motor[b]				
	Visual Motor Integration	90.78 (12.81)	92.39 (10.72)	0.48
	Motor Coordination	90.56 (6.75)	92.58 (8.43)	0.95
		DCD		
n		23	22	
Age (months)		117.61 (10.47)	117.55 (9.97)	0.02
Non-verbal IQ[a]		8.52 (2.63)	9.00 (2.79)	0.59
Literacy[b]				
	Word Spelling	104.57 (15.51)	112.41 (12.98)	1.84
	Word Reading	101.09 (13.22)	106.95 (9.95)	1.71
Motor[b]				
	Visual Motor Integration	82.13 (10.89)	94.48 (10.20)	3.87***
	Motor Coordination	75.57 (9.95)	91.55 (8.97)	5.65***
		Comorbid Dyslexia and DCD		
n		15	15	
Age (months)		118.93 (13.19)	118.8 (11.83)	0.03
Non-verbal IQ[a]		6.60 (2.59)	8.33 (1.80)	2.13*
Literacy[b]				
	Word Spelling	74.27 (9.00)	108.87 (13.86)	8.11***
	Word Reading	83.07 (6.41)	104.53 (10.59)	6.72***
Motor[b]				
	Visual Motor Integration	73.60 (9.63)	94.86 (8.32)	6.34***
	Motor Coordination	78.13 (11.34)	90.4 (6.17)	3.68***

Note. Standard deviations are reported in the parenthesis. Non-verbal IQ was measured using the
WISC-IV Block Design (Wechsler et al., 2004)
[a]Scaled scores. [b]Standard scores.
p < .05 **p* < .01, ****p* < .001.

7.2.2. Design and Procedure

In this study, a modified two-session training paradigm after Taylor et al. (2012) was used. In the first phase, children were trained by copying novel characters. Post-training learning was assessed one to three days later, also by copying novel characters. In addition to copying novel characters, we also asked children to copy alphabet letters in order to provide context of highly familiar letters from which to compare novel characters with. Children completed both phases individually either in a quiet area of their school or in a testing room at Bangor University. In all instances, children wrote their responses on a piece of paper attached to a digitising tablet. The digitising tablet was used to capture graphomotor behaviour, specifically pausing behaviours whilst writing.

7.2.2.1. Session one. During Session 1 children completed the baseline, training, and alphabet letter copying tasks.

7.2.2.1.1. Baseline for novel characters. Pre-training measurements of children's copying of each of the 25 novel characters were taken. Children were told they were going to see a special letter on the screen which would not be familiar to them and they should copy the letter onto the paper in front of them – affixed to the top of a digitising tablet – as quickly as they could in their normal handwriting. Once they had finished copying the letter they were to press their pen tip onto a green rectangle at the bottom of the paper to signal they had finished the trial. Pressing their pen into the green rectangle automatically triggered the end of the trial. Each trial began with a 400 ms central fixation cross followed by the appearance of a novel character (size 32 font) presented in the centre of the screen (see Figure 7.1). The order of the characters was randomised for each child and the characters remained on the screen for the duration of the trial.

7.2.2.1.2. Training. Immediately following the baseline measurements, children copied

the same novel characters grouped in strings of five different characters over eight blocks. In each block, five strings of five-characters were presented across the middle of the screen (see Figure 7.1.). This meant that each character was copied once per block. Children were reminded to copy all the 'funny letters' as accurately and as quickly as they could in their normal handwriting but without joining the letters up. Each block was self-paced and ended when the child pressed their pen onto a green rectangle on their page to signal they had finished the trial.

The rationale for string size was based on a search of the Children's Printed Word Database (CPWD; Stuart, Masterson, Dixon, & Quinlan, 1993) that revealed five letter words to be the average length of a word in English – with a frequency greater than 40 words per million – that children aged 7 to 8 years old are exposed to. Within each trial, the five strings each containing five characters were presented simultaneously to maintain the participants' focus and to reduce the overall session time. The strings were constructed to ensure the total visual orientation and motor complexity of the characters within each string were as closely balanced as possible (see Stimuli). Within strings, characters were pseudo-randomly assigned a position to ensure no character was copied from the same character position more than twice between blocks. Within each block, string position was also pseudo-randomised to ensure no string was copied from the same position more than twice.

7.2.2.1.3. Alphabet copy. Measures of alphabet writing were taken following training. Using the same novel character pre-training procedure. Children copied individual alphabet letters from the screen onto the lined paper. Each trial began with a 400 ms central fixation cross followed by appearance of the alphabet letter (size 32 font) presented in the centre of the screen (see Figure 7.1). The order of the letters was randomised, and they remained on the screen for the duration of each trial which ended when children pressed their pen into the green rectangle on

their paper.

7.2.2.2. Session two. Children completed Session 2, one to three days after Session 1 when copying accuracy and handwriting parameters were re-assessed for novel characters and alphabet letters. In both conditions, each item was preceded by a 400ms fixation cross, presented centrally on the screen (font size 32) and remained until the child signalled they had completed the trial by pressing their pen tip onto the green rectangle at the bottom of the paper.

7.2.2.2.1. Novel characters. Using the same individual character/letter copying procedure as Session 1, children copied the individual novel characters from a laptop screen onto lined paper.

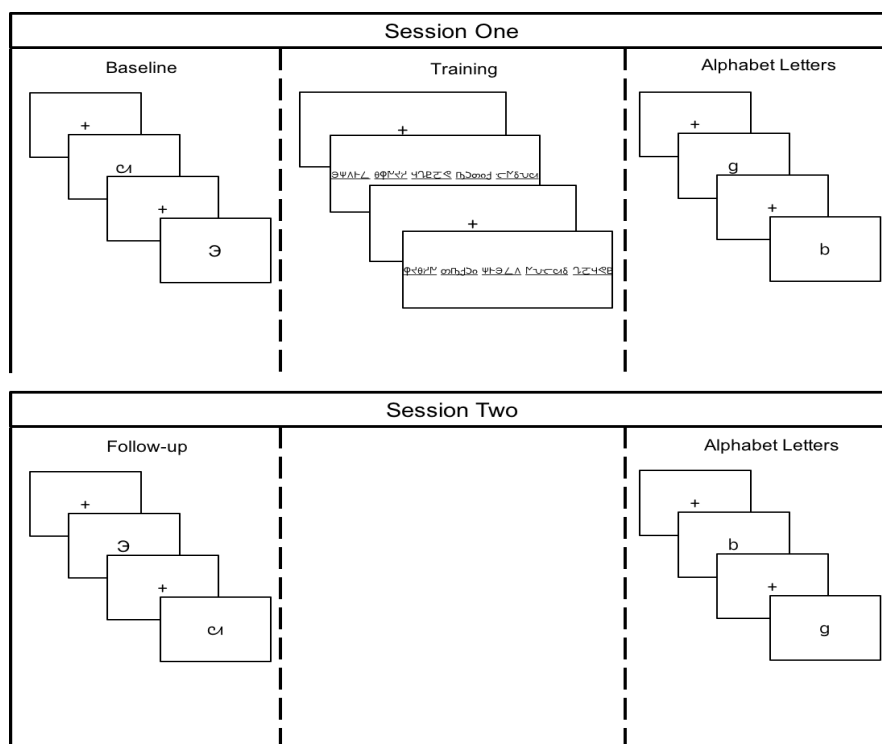*7.2.2.2.2. Alphabet Letters.* In a second condition, children copied individual alphabet letters.

Figure 7.1. Experimental novel orthographic character learning procedure. During Session 1, children copied novel characters individually (baseline), then in strings (training), followed by

individually presented alphabet letters. In Session 2, one to three days later, children copied

novel characters (follow-up) and then alphabet letters.

7.2.3. Stimuli

A bank of novel characters was created by extracting letters from a number of non-Latin

orthographies which were unfamiliar to children (e.g., aboriginal Canadian). Each character and

lowercase alphabet letter was transformed into Yuanti SC font. Yuanti SC font was used for all

stimuli in this study as it fitted the following criteria (a) The font's curves and straight lines were

consistent across novel characters from different orthographies, (b) the construction of the typed

alphabet letters and novel characters closely approximated handwritten construction of the

letters/characters (e.g., $<a>$ instead of an $<a>$).

The visual orientation and motor complexity of novel characters and lowercase alphabet

letter was then rated by the candidate. After rating, novel characters were matched with each

alphabet letter as closely as possible on their visual orientation and motor complexity. The letter

$<l>$ could not be matched with a novel character and so was not included as a stimulus in this

study. This procedure resulted in 25 alphabet letters and 25 novel characters.

7.2.3.1. Orientation. To determine the character's orientation, we followed Treiman and

Kessler's (2011) definition. Characters with the hasta clearly positioned on the right of the coda

were categorised as a b-type character, whilst characters with the hasta clearly positioned on the

left of the coda was categorised as a d-type character. Characters without a clear hasta-coda

combination (e.g., $<o>$) were categorised as n-type characters.

To ensure accuracy and consistency of the ratings, we asked 30 adults (*M* age = 26.5

years, 80% female) to rate the visual orientation of both the alphabet letter and novel characters.

The full breakdown of the results from the rating study are presented in Appendix E. There was

96% and 92% agreement between the candidate's ratings of orientation and those made by the volunteer raters for alphabet letters and novel characters respectively. Where there was disagreement between the ratings, we used the ratings given by the volunteers.

7.2.3.2. Strokes. Numerous methods have been used to measure the number of component strokes required to form characters. The methods range from simply partitioning each letter according to the minimum number of strokes needed to produce a recognisable letter (e.g., Treiman & Kessler, 2011) to more complex online methods which involve segmenting characters according to where the pen slows down (tangential velocity minima; e.g., Kandel & Spinelli, 2010). The latter technique is precise and is very information rich. For example, it accounts for the decreases in velocity when forming curved and rotational strokes. However, this technique is time consuming and excess to the requirement of this study and so we used a modified version of Treiman and Kessler's (2011) criteria. Following these modified criteria, the number of strokes of characters/letters was measured by counting the number of separate straight and curved lines required to form the character (see Figure 7.2).

To validate the adapted measure of number of strokes, we asked the same 30 volunteer raters who judged the visual orientation of the characters/letters to also rate the number of strokes in each of the characters/letters (see Appendix E). There was low agreement between the candidate's and volunteer's ratings for alphabet letters (68%) and novel characters (84%). The source of the low agreement was restricted to letters/characters that featured a curve at the top or bottom of the stem (e.g., < *y* >, < *f* >). The candidate distinguished between the curve and the straight line of the stem, but the volunteer raters did not, particularly for the highly familiar alphabet letters.

To clarify which ratings were the most valid, the candidate's and the volunteer's complexity ratings were correlated with the pausing duration within each letter/character (intra-character pausing duration). Intra-character pausing duration was used as a proxy of fluency, based on the assumption that letters/characters with a greater number of strokes will lead to longer and more frequent pausing, reducing the overall fluency. The candidate's motor complexity ratings correlated highly with the intra-character pausing durations of alphabet letters, $r = .51$, $p = .01$ and novel characters, $r = .63$, $p < .001$, however, the volunteer's ratings did not correlate with intra-character pausing durations as well (alphabet letters, $r = .31$, $p = .136$; novel characters' $r = .52$, $p = .008$). Accordingly, the candidate's ratings of motor complexity were chosen because these ratings correlated more strongly with pausing behaviours, an objective proxy measure of complexity.
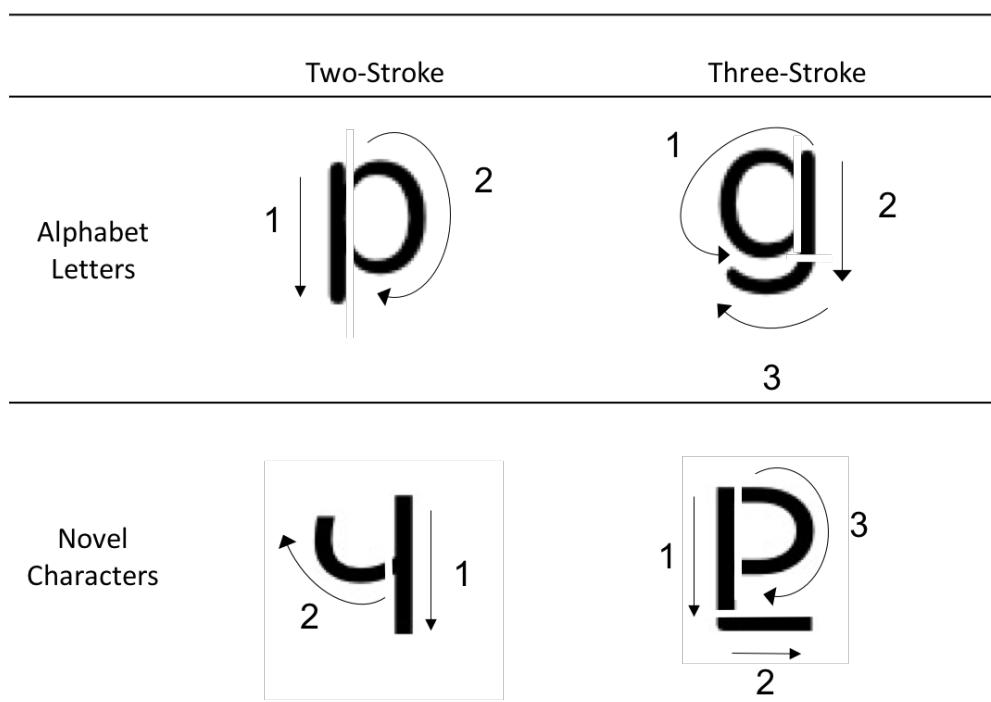

Figure 7.2. Example segmentation of procedure alphabet letters (top panel) and novel characters (bottom panel). Letters/characters were segmented into elements (straight and curved lines).

7.2.4. Apparatus

Simultaneous administration of the experiment and recording of pen tracking data was undertaken using Eye and Pen 2.0 (Alamargot et al., 2006). The programme was run on a Dell Precision M4800 15-inch laptop connected to a medium Wacom Intuos Pro digitising tablet (sampling area: 224 x 140mm) sampling at 200Hz. The digitising tablet was positioned horizontally with lined A4 paper attached over the top of the pad to mimic the way children would write in their exercise books. Children wrote onto the paper using a compatible Wacom Inking pen which acts as a digitising stylus as well as leaving a visible ink trace on the paper. Participants were briefed to hold and write with the 'special pen' as if it were a normal ballpoint pen. All children had experience with writing using this set up from previous studies.

7.2.5. Data Analysis

The primary measure in this study was pausing durations. Pauses are defined as a period of inactivity during writing (Olive, 2010). They index overall handwriting fluency whereby longer pauses increase the total time to form a letter (Paz-Villagrán, Danna, & Velay, 2014). Pausing behaviours were chosen for this study as they account for discontinuity in a trace and discriminate between children with and without dyslexia and/or DCD (e.g., Prunty et al., 2013; Sumner et al., 2013; Study 3). Pauses can be subdivided into pre-writing pauses and intra-character pauses. Pre-writing pauses are akin to onset latency and account for the time between the stimulus appearing on the screen and the first touch of the child's pen onto the paper. They are related to planning, formulating, and retrieving information (Chenu, Pellegrino, Jisa, & Fayol, 2014). Intra-character pauses account for pausing that takes place whilst forming the letter and are related to monitoring and repairing (Chenu et al., 2014).

7.2.5.1. Data extraction. Data were extracted for each letter/character individually using Eye and Pen 2.0 (Alamargot et al., 2006). Pausing durations were set at a low threshold of 30ms. This low threshold was set to capture both lower and higher level motor and cognitive processes (e.g., Olive & Kellogg, 2002). As well as extracting pausing durations, the accuracy of each letter/character was scored using modified criteria from the Letter Formation dimension of the SaHLT (Caravolas & Downing, in prep.). Responses were scored using a binary correct/incorrect scale. They were scored as correct if they were: (a) recognisable from the model, (b) included all the strokes, (c) strokes were formed correctly, (d) the letter was not reversed. Accuracy was scored by the candidate. To ensure reliability, a trained research assistant scored a randomly selected 10% of the sample. The inter-rater reliability between the candidate's and the research assistant's scores for alphabet letters and novel characters was assessed using two-way random effects intra-class correlations (r_{ICC}). The inter-rater reliability for alphabet letters was good, r_{ICC} = .89, 95% CI [.51, .97] and for novel characters was excellent, r_{ICC} = .96, 95% CI [.84, .99].

7.2.5.2. Statistical analyses. To balance the number of letters/characters with different orientations and number of strokes, performance was analysed on a subset of the letters and characters. Characters were selected if they had either a b-type or d-type orientation and were composed of two or three strokes as these were the most common visuomotor properties of alphabet letters (see Appendix F for breakdown of letters/characters by visuomotor properties). Analyses were conducted on both accuracy and pausing data. In each case, alphabet letters were analysed first to provide context of highly familiar letters for a contrast with the analysis of pausing behaviours on novel characters, the main focus of this study. Due to the large number of separate analyses, the specifics of each analysis are described in the results for clarity.

7.3. Results

This study was concerned with investigating whether letter formation difficulties present in dyslexia and DCD could be explained, in part, by impairments in learning motor programmes. The aims were addressed in the following analyses by probing whether children with dyslexia and/or DCD made longer pauses and were therefore less fluent than typically developing children after being trained on novel characters varying in visuomotor complexity. Further evidence of impairments in learning motor programmes would be apparent if children with dyslexia and/or DCD but not typical developing children's pausing durations were affected by the visuomotoric complexity of the characters. Prior to addressing these questions using the primary measure of pausing behaviours, accuracy data were analysed as a function of group and characters' visuomotor properties.

7.3.1. Accuracy

Accuracy was high across all groups and conditions for both alphabet letters and novel characters. Due to the high degree of accuracy the data were heavily skewed which prevented the use of parametric tests and so non-parametric alternatives were used to examine performance between groups and the visuomotor properties of the characters. Wilcoxon Signed-rank (Z) tests were used to assess differences in repeated measures. For between subjects, Kruskal-Wallis (χ^2) tests were used. In cases where multiple comparisons were made, Bonferroni corrections were applied. Significant Kruskal-Wallis tests were followed up using Dunn tests with Bonferroni corrections. Analyses examined accuracy when (a) copying alphabet letters, (b) copying novel characters between baseline and follow-up, and (c) copying novel character strings during training.

7.3.1.1. Alphabet letters. Children copied single alphabet letters from the screen in Session 1 and again in Session 2. A coding error in the experiment meant no data from alphabet letter copying at Session 1 was recorded from 33 children. Of those children, ten were in the dyslexia group, eight in the DCD group, three in the comorbid dyslexia and DCD group, and twelve in the comorbid group. Due to the large amount of missing data, data were aggregated across groups to examine accuracy between the sessions. Children were similarly accurate in Session 1 (Mdn = .93) and Session 2 (Mdn = .93) and this difference was not statistically significant, Z = .24, p = .810.

As there was no difference in accuracy between sessions, accuracy was compared between groups and the visuomotoric properties of characters from Session 2 which included data from all participants. At Session 2, median accuracy of alphabet letters significantly differed between groups, $\chi^2(3)$ = 28.04, p < .001. Children with DCD (Mdn = .85) were significantly less accurate at copying alphabet letters than typically developing children (Mdn = 1; p < .001), children with dyslexia (Mdn = .94; p = .006) and children with comorbid dyslexia and DCD (Mdn = .94; p = .048). No other comparisons between groups reached statistical significance. Median accuracy did not differ significantly between letters with different orientations or number of strokes.

7.3.1.2. Novel characters. Children copied novel characters at baseline and follow-up. During training, children also copied the same characters in strings. Accuracy was higher when copying individual novel characters than when copying the characters in strings, reflecting the increased difficulty of copying strings. Group and visuomotor effects on accuracy were analysed at baseline and at follow up to ascertain whether accuracy changed as a result of training. Further

analysis of accuracy during training were run to examine whether accuracy increased during training.

7.3.1.2.1. Baseline and follow-up. At baseline, group's accuracy differed significantly, $\chi^2(3) = 11.47$, $p = .009$. Children with DCD (*Mdn* = .85) were significantly less accurate than typically developing children (*Mdn* = .93; $p = .003$). Children with dyslexia (*Mdn* = .85) and children with comorbid dyslexia and DCD (*Mdn* = .83) were also less accurate than typically developing children, however the difference between these groups did not reach statistical significance. Differences in accuracy between any of the disorder groups failed to reach statistical significance.

At follow up, accuracy differed significantly by group again, $\chi^2(3) = 18.57$, $p < .001$. Children with DCD (*Mdn* = .81) and comorbid dyslexia and DCD (*Mdn* = .73) were significantly less accurate than typically developing children (*Mdn* = .93; *p*s < .01) and children with dyslexia-only (*Mdn* = .92; *p*s < .05). Differences between typically developing children and children with dyslexia and between children with singular and comorbid DCD did not reach statistical significance. Analyses within groups between baseline and follow-up were non-significant. Moreover, differences between characters with different visuomotor properties failed to reach statistical significance.

7.3.1.2.2. Training. During training, accuracy decreased in all groups. In typically developing children, there was a non-significant decrease in accuracy between Blocks 1 (*Mdn* = .85) and 8 (*Mdn* = .83), $Z = 1.45$, $p = .147$. Children with dyslexia were less accurate than typically developing children, and there was also a (non-significant) decrease in accuracy between Blocks 1 (*Mdn* = .77) and 8 (*Mdn* = .65), $Z = 0.95$, $p = .341$. Children with DCD made a similar number of errors to children with dyslexia, and their median accuracy did not differ

between Block 1 (*Mdn* = .73) and Block 8 (*Mdn* = .64), after applying Bonferroni corrections for multiple comparisons, Z = 2.36, p > .05. Children with comorbid dyslexia and DCD had the lowest accuracy in Bock 1 (*Mdn* = .69) and Block 8 (*Mdn* = .58). The comorbid group's accuracy did not differ significantly between Blocks 1 and 8, Z = 0.40, p = .691. Median accuracy did not differ significantly between training blocks according to the visuomotor properties of the characters. Following training, children completed a short visual recognition task which revealed all children – regardless of group – were able to correctly identify novel characters they had copied during training (see Appendix G).

The finding that accuracy decreased, albeit non-significantly, is somewhat counter to expectations. A possible explanation for this is fatigue. Copying 25 novel characters in a row is an intensive exercise and requires a high level of concentration. Copying the same 25 novel characters in a row over eight consecutive blocks means that this level of concentration must be sustained for quite some time. It could be that the drop-in accuracy reflects difficulties in sustaining concentration for such a prolonged period of time.

7.3.2. Pausing Durations

Pausing behaviours were measured using the digitising tablet and Eye and Pen software. In the following analyses differences between groups and visuomotor properties of characters were examined on pausing durations of correctly formed letters/characters only. Pausing behaviour was only examined on correctly formed characters to account for the group differences in accuracy reported in the preceding section. Outliers were Winsorized to within the 10th and 90th percentiles and data were checked to ensure they met the assumptions for parametric analyses. Where data were non-normally distributed, transformations were used to improve the distributions.

Pre-writing and intra-character pause durations were analysed separately for alphabet letters and novel characters. Analyses of alphabet letter formation examined fixed effects of group and visuomotor properties of characters at post training. Analyses of novel character formation examined fixed effects of time, group, and visuomotor properties at baseline and follow-up. Further analyses of novel characters also examined performance during training via string copying. In each analysis, comparisons were made using analyses of variance (ANOVA) models. The designs differed according to the analyses and so are described with each analysis reported. In each analysis involving comparisons with the comorbid dyslexia and DCD group, WISC Block Design scores were entered as a covariate to account for significantly lower NVIQ scores. In all cases the covariate NVIQ was non-significant and so the analyses without NVIQ as a covariate are reported.

7.3.2.1. Alphabet Letters. Due to the experimental error described earlier, differences between groups and visuomotor properties of the characters on pre-writing and intra-character pausing durations were tested on data from Session 2 only.

7.3.2.1.1. Pre-writing pause durations. Initial analyses revealed pre-writing pause duration did not differ by the letter's visuomotor properties and so the data was aggregated across these properties to examine group differences. Subsequently, group differences were examined using a one-way ANOVA. Figure 7.3 depicts the mean pre-writing pause durations of each group when copying alphabet letters. There were large differences between group's pre-writing pause durations, $F(3, 94) = 8.57$, $p < .001$, $\eta_\rho^2 = .67$. Children with dyslexia-only and comorbid dyslexia and DCD took longer than typically developing children to initiate letter formation ($ps < .05$). Children with DCD also took longer to begin writing than typically

developing children although this difference failed to reach statistical significance. Pre-writing

pauses did not differ significantly between the disorder groups.
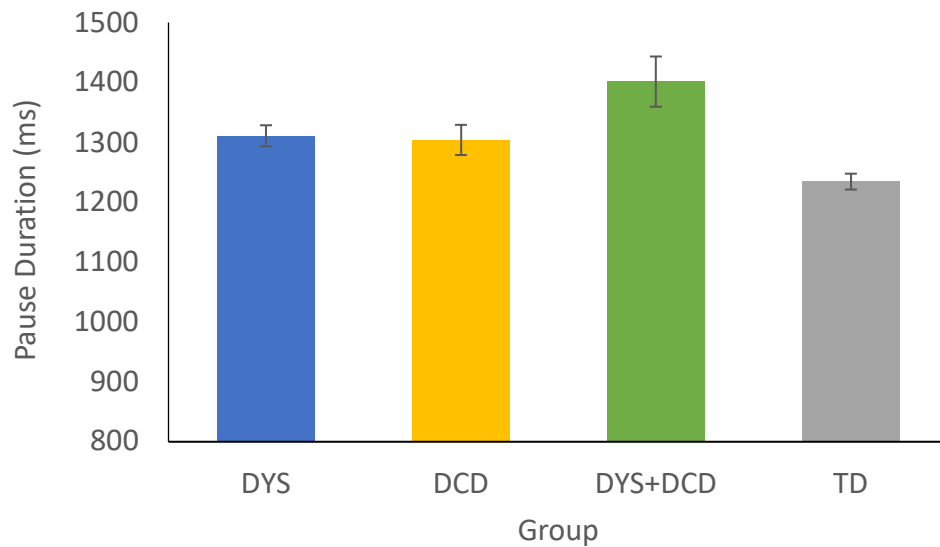
Figure 7.3. Mean pre-writing pause durations of alphabet letters as a function of group. DYS =

dyslexia. DYS+DCD = comorbid dyslexia and DCD. TD = typically developing children

(TD). Error bars represent standard error of the mean.

    7.3.2.1.2. Intra-character pause durations. Initial 2 (group: disorder vs control) x 2

(orientation: b-type vs d-type) x 2 (stroke: two-stroke vs three-stroke) ANOVAs for each

disorder group and their respective controls failed to find any significant group effects or

interactions. Each analysis did reveal consistent effects of visuomotor properties on intra-

character pausing durations. To better understand how intra-character pause durations differed by

visuomotor properties the data were aggregated across letters for an item analysis. A 2

(orientation: b-type vs d-type) x 2 (stroke: two-stroke vs three-stroke) ANCOVA, co-varying for

letter frequency, examined differences in intra-character pause durations. Proxy letter frequency

estimates were derived by averaging the frequency of words which contained the letter from the

CPWD (Masterson, Stuart, Dixon, & Lovejoy, 2010). The frequency covariate was non-significant and so a second model was run without frequency. Children made longer pauses when copying more complex, three-stroke letters ($M = 282.33$, $SD = 142.05$) than when copying two-stroke letters ($M = 142.48$, $SD = 94.12$), $F(1, 10) = 5.25$, $p = .045$, $\eta_\rho^2 = .34$. Children's pausing durations did not significantly differ between b-type ($M = 223.25$, $SD = 135.54$) or d-type ($M = 182.02$, $SD = 138.00$) orientations. There was no significant interaction on pausing durations between orientation and number of strokes.

7.3.2.2. Novel characters. This analysis addressed the key question of this study. That is, whether children with dyslexia and/or DCD have impairments in learning letter-like motor programmes. To address this question, groups and their matched controls were analysed separately (see Table 7.1) in order to negate homogeneity of variance issues from large differences in sample sizes between the groups. Previous analyses conducted in this thesis – using less complex designs – were more robust for differences in sample sizes and we controlled for large differences by using sample weights. However, weights cannot be applied in designs with repeated measures, as is the case here. An added benefit of using separate analyses is that they allow for close matching between typical developing children and each disordered group. Group and visuomotor property differences were tested using 2 (group: disordered vs typically developing) x 2 (time: baseline vs follow-up) x 2 (orientation: b-type vs d-type) x 2 (stroke: two-stroke vs three-stroke) mixed ANOVAs.

7.3.2.2.1. Novel character pre-writing pause durations. The data were positively skewed and so were normalised using inverse transformations. Analyses of the transformed and non-transformed data yielded the same results and so the non-transformed data are discussed here. Separate analyses of pre-writing pausing durations for each disorder group and their respective

controls failed to find any significant group effects or interactions. Each analysis did however reveal consistent effects of time and visuomotor properties on pre-writing pause durations.

To examine differences in the time it took for children to begin writing as a function of visuomotor properties of characters, the data were aggregated across groups and analysed using a three-way, 2 (time: baseline vs follow-up) x 2 (orientation: b-type vs d-type) x 2 (stroke: two-stroke vs three-stroke) repeated measures ANOVA. The mean pre-writing pause durations as a function of character's visuomotor properties are reported in Figure 7.4. Children made shorter pauses before writing at follow-up than at baseline, $F(1, 96) = 198.65$, $p < .001$, $\eta_\rho^2 = .67$. Pauses were shorter for b-type characters than d-type characters, $F(1, 96) = 15.05$, $p < .001$, $\eta_\rho^2 = .13$. Pauses were also marginally shorter for two-stroke characters than three-stroke characters, $F(1, 96) = 6.56$, $p = .062$, $\eta_\rho^2 = .04$.

In addition to the main effects, there was also a significant Orientation x Stroke interaction, $F(1, 480) = 29.36$, $p < .001$, $\eta_\rho^2 = .06$. Simple effects analysis revealed when copying b-type characters, pre-writing pausing durations were shorter when the characters were composed of two-strokes ($p < .001$) but not three-strokes. Finally, there was also a significant three-way, Time x Orientation x Stroke, interaction $F(1, 480) = 11.96$, $p < .001$, $\eta_\rho^2 = .02$. Simple effects analysis revealed when copying b-type characters, pre-writing pausing durations were shorter when the characters were composed of two-strokes at baseline ($ps < .001$). No other character of different visuomotor properties differed from each other.
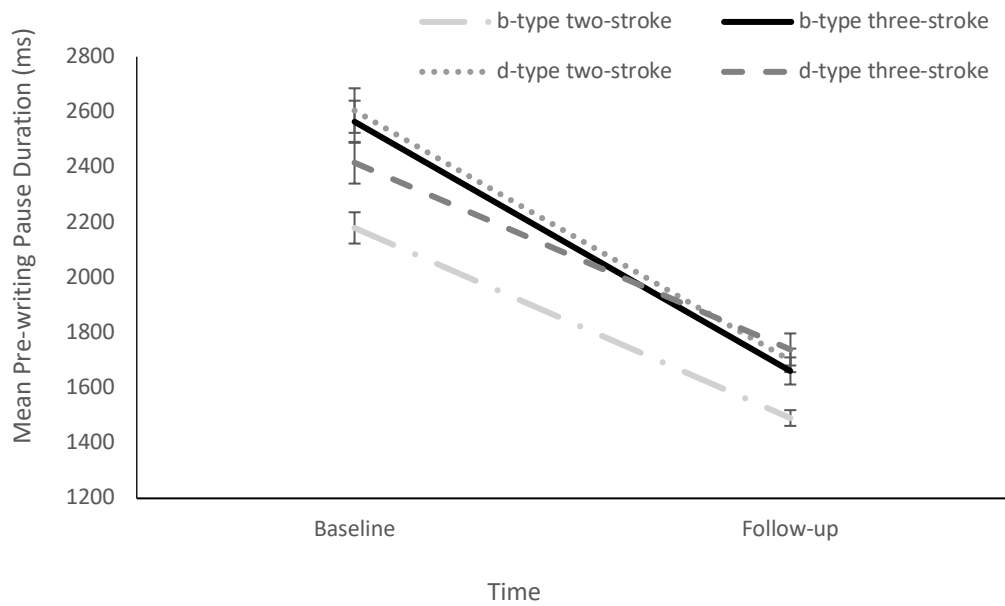
Figure 7.4. Mean pre-writing pause durations of novel characters as a function of the character's visuomotor complexity at baseline and follow-up. Error bars represent standard error of the mean.

7.3.2.2.2. Novel character intra-character pausing durations. The pausing durations within novel characters were highly positively skewed. The distributions of the data were improved using log-transformations and the log-transformed data was analysed using the separate four-way, 2 (group: disordered vs typically developing) x 2 (time: baseline vs follow-up) x 2 (orientation: b-type vs d-type) x 2 (stroke: two-stroke vs three-stroke) mixed ANOVAs described earlier.

7.3.2.2.2.1. Dyslexia. Mean pausing durations of children with dyslexia and matched controls are shown in Figure 7.5a. As expected, there was a large decrease in pausing between baseline and follow-up, $F(1, 49) = 210.75$, $p < .001$, $\eta_\rho^2 = .81$. On differences in visuomotor properties, children paused for less time when copying b-type characters, $F(1, 49) = 26.67$, $p <$

.001, η^2_ρ = .35, and when copying two-stroke characters, $F(1, 49) = 110.02$, $p < .001$, η^2_ρ = .69. There was no significant main effect of group, however, there was a significant Group x Time interaction, $F(1, 245) = 7.48$, $p = .009$, η^2_ρ = .13. Children with dyslexia made longer pauses than controls whilst copying characters at follow-up ($p = .011$), but not at baseline. Interactions between the visuomotor properties of characters with time and/or group failed to reach statistical significance.

7.3.2.2.2.2. DCD. Intra-character pausing durations between groups and visuomotor properties of characters before and after training are reported in Figure 7.5b. Again, children made large decreases in pauses between baseline and follow-up, $F(1, 43) = 128.81$, $p < .001$, η^2_ρ = .75. Pauses were shorter whilst copying characters with a b-type orientation, $F(1, 43) = 6.90$, $p = .012$, η^2_ρ = .14, and characters with two-strokes, $F(1, 43) = 43.29$, $p < .001$, η^2_ρ = .5. Similarly, to the analysis of dyslexic group, there was a significant Time x Group interaction, $F(1, 215) = 9.85$, $p = .003$, η^2_ρ = .19. Children with DCD made longer pauses than controls whilst copying characters at follow-up ($p = .014$), but not at baseline. No other interactions between the visuomotor properties of characters with time and/or group reached statistical significance. The pausing durations of children with DCD did not differ significantly from those of children with singular dyslexia, $t(48) = 0.44$, $p = .66$, $d = .13$.

7.3.2.2.2.3. Comorbid dyslexia and DCD. Pausing durations whilst copying characters for children with comorbid dyslexia and DCD and matched typically developing controls are displayed in Figure 7.5c. In this analysis also, there was a large decrease in pausing durations between baseline and follow-up, $F(1, 28) = 93.70$, $p < .001$, η^2_ρ = .77. On visuomotor properties, pauses were shorter for b-type characters, $F(1, 28) = 7.62$, $p = .01$, η^2_ρ = .21, and characters with two-strokes, $F(1, 28) = 46.77$, $p < .001$, η^2_ρ = .63. Children with comorbid dyslexia and DCD

paused for longer than typically developing controls, $F(1, 28) = 7.84$, $p = .009$, $\eta_\rho^2 = .22$. The time by group interaction present in the singular disorder analyses did not reach significance here. Therefore, children with comorbid dyslexia and DCD paused for longer than typically developing controls at both baseline and at follow-up. Furthermore, no other interactions reached significance. Contrasts between the disorder groups at follow-up revealed pausing durations did not differ significantly between the comorbid group and the singular dyslexia group, $t(40) = 0.17$, $p = .86$, $d = .06$ or the singular DCD group, $t(36) = 0.15$, $p = .88$, $d = .05$.
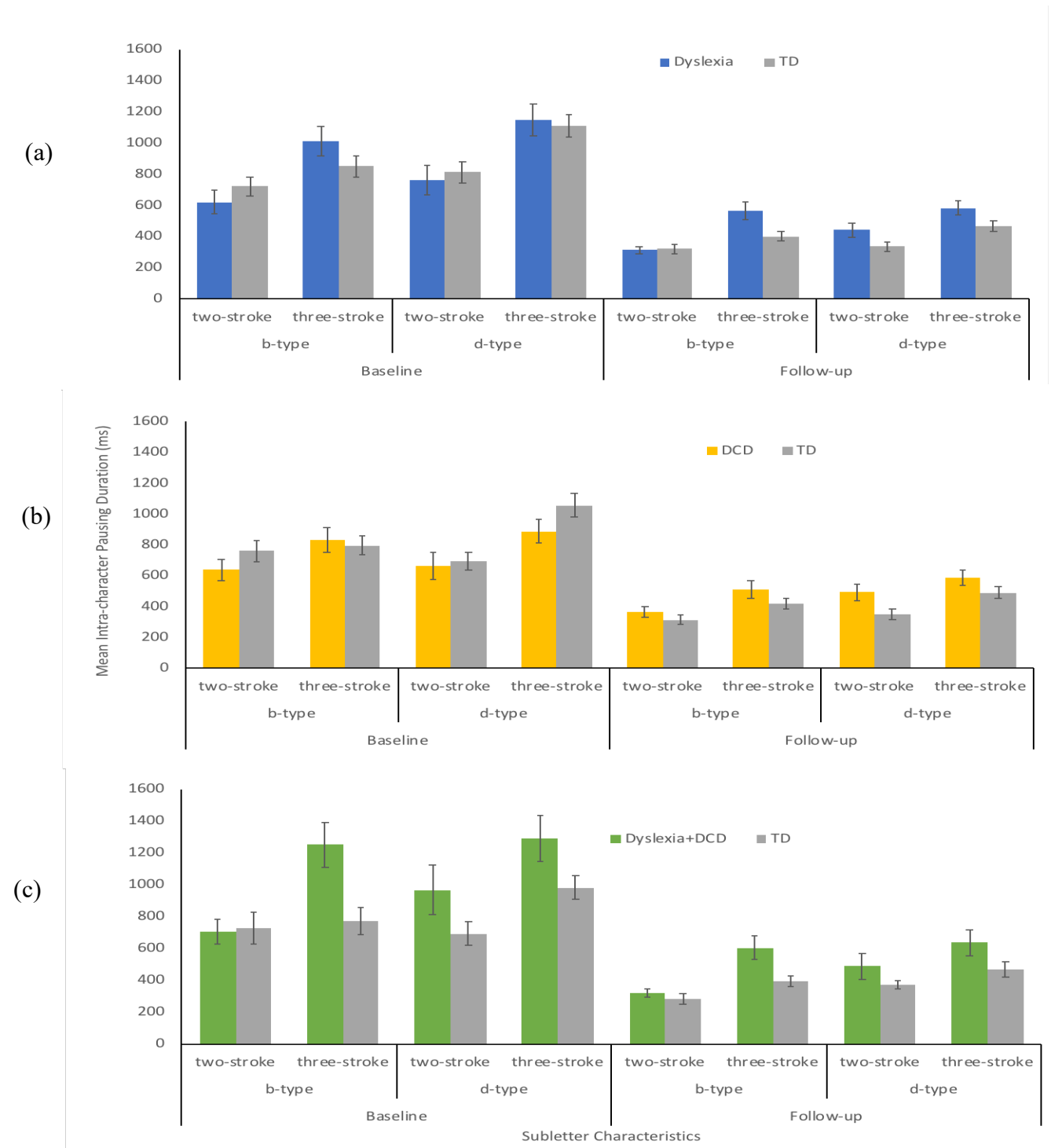
Figure 7.5. Mean intra-character pausing durations as a function of character's visuomotor properties at baseline and follow-up for children with (a) dyslexia, (b) DCD, and (c) comorbid dyslexia and DCD and their respective matched controls. Error bars represent standard error.

7.3.3. Group Differences in Pausing Behaviours During Training

The previous analysis revealed children dyslexia and DCD paused for longer when writing novel characters. The nature of this pausing behaviour is interesting. Children with dyslexia-only and DCD-only paused for longer than controls after training but not before. This finding suggests that impairments in learning motor programmes are present in both dyslexia and DCD. However, the previous analyses do not detail whether problems in learning motor programmes begin during the initial encoding stages of learning (e.g., training) or during consolidation (between training and follow-up).

To examine whether impairments in learning motor programmes lie in the encoding, performance was also compared during training. To do so, a training value was calculated for each child by subtracting the intra-character pause durations of correctly copied characters in Block 8 from the correctly copied characters in Block 1. In cases where participants did not copy any items in Blocks 1 or 8 correctly, the values were subtracted from Blocks 2 and/or 7. This training value measures the magnitude of the training effect for each child. Negative values indicate that pausing decreased during training and therefore letter formation became more efficient. Thus, a large negative value would indicate a large increase in fluency and a small negative value would indicate a small increase in fluency.

Mean training values for each group are reported in Figure 7.6. On average, all groups decreased the time they spent pausing during training. Children with dyslexia (singular and comorbid) made smaller decreases in pausing durations during training, however, group differences in training values failed to reach statistical significance, $F(3, 94) = 0.49$, $p = .69$, $\eta_\rho^2 = .06$.
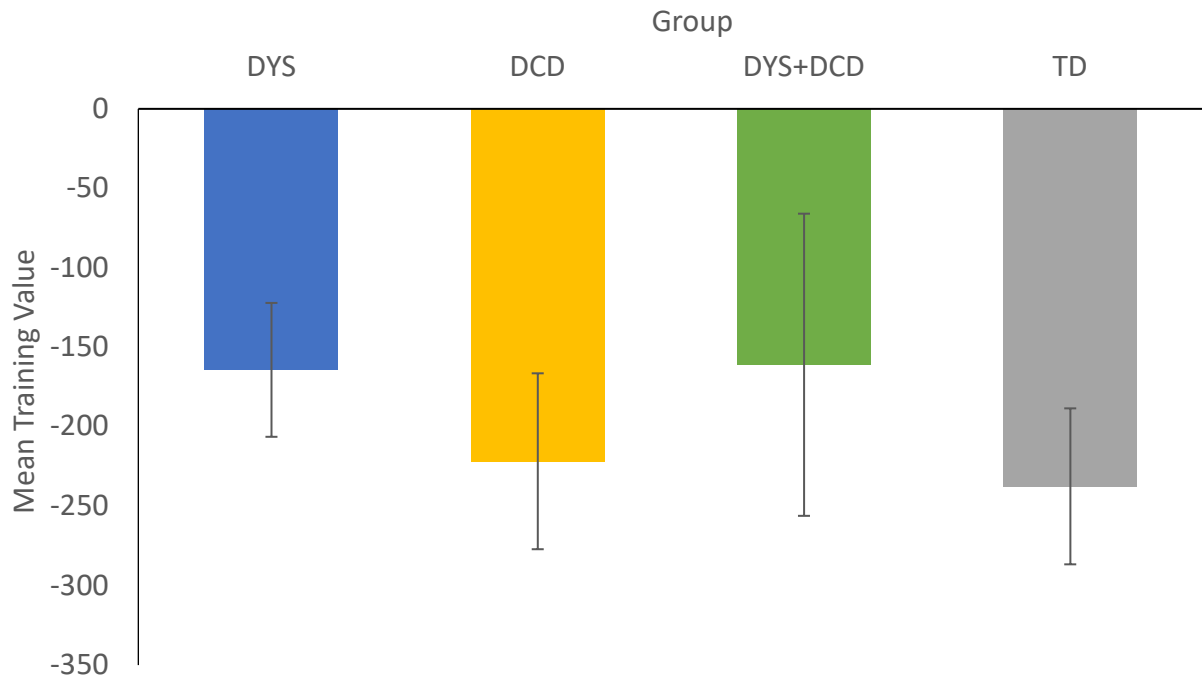
Figure 7.6. Mean training values (decrease in intra-character pause durations during training) as a function of group. DYS = dyslexia. DYS+DCD = comorbid dyslexia and DCD. TD = typically developing children. Error bars represent standard error.

7.3.4. Relationships Between Pausing Behaviours and Handwriting Related Skills

In this final analysis, relationships between pausing durations and handwriting related skills (phonological, literacy, perceptuomotor, and executive function) were examined. To do so, Pearson's correlations between pausing durations and factor score estimates derived from the MIMIC model in Study 2 were run separately for the dyslexia-only, DCD-only, and typically developing groups. Correlations were not run for the comorbid dyslexia and DCD group due to its small size.

7.3.4.1. Dyslexia. Children who made longer pre-writing pauses when copying alphabet letters also made longer pre-writing pauses when copying novel characters at baseline ($r = .46$, $p = .02$) and at follow-up ($r = .57$, $p < .001$). Pre-writing pause durations whilst copying novel

characters (baseline and follow-up) and alphabet letters were not significantly related to any of

the factor score estimates in this group. However, intra-character pauses did correlate with

perceptuomotor and executive function factor score estimates. Children with dyslexia who made

longer intra-character pauses when copying novel characters at follow-up had lower

perceptuomotor ($r = -.39$, $p = .04$) and executive function ($r = -.35$, $p = .06$) factor score

estimates.

7.3.4.2. DCD. Children with DCD who made longer pre-writing pauses when copying

novel characters made longer pre-writing pauses when copying alphabet letters at follow-up ($r =$

$.47$, $p = .018$). Intra-character, but not pre-writing, pauses significantly correlated with factor

score estimates of perceptuomotor skills in this group. Children with lower perceptuomotor skills

made longer intra-character pauses when copying novel characters at baseline ($r = -.52$, $p = .016$)

and marginally so at follow-up ($r = -.47$, $p = .058$).

7.3.4.3. Typically developing. Typically developing children who paused for longer

before writing at follow-up also paused for longer before copying alphabet letters ($r = .40$, $p =$

$.02$). A similar pattern of correlations was found for the intra-character pause durations. Children

who made longer intra-character pause durations when copying novel characters also made

longer pause durations when copying alphabet letters at baseline ($r = .36$, $p = .04$) and at follow-

up ($r = .36$, $p = .05$). Typically developing children's pause durations were not significantly

related to their factor score estimates. As in Study 3, comparisons of Pearson's correlation sizes

between groups failed to reach statistical significance in all cases which likely reflects a lack of

power in the analyses.

7.4. Discussion

This study was concerned with investigating whether letter formation difficulties apparent in both dyslexia and DCD (Study 3) could be explained, in part, by impairments in learning the motor programmes of letters. Learning motor programmes is difficult and prior to consolidating them, children are sensitive to the visuomotor properties of characters. It was hypothesised that if children with dyslexia and/or DCD had impairments in learning motor programmes, they would pause for longer, and therefore be less fluent, than typically developing children after training. It was further hypothesised that children with dyslexia and/or DCD would remain sensitive to the visuomotor properties of letters for longer and make shorter pause durations when copying visuomotorically less complex (e.g., b-type orientation with fewer strokes) characters after training, but typically developing children would not. The results were consistent with the first hypothesis but not the second.

On accuracy, children with DCD-only were less accurate than typically developing children when copying alphabet letters and novel characters. Accuracy was not affected by dyslexia status (singular or comorbid) or by the visuomotor properties of characters. An unexpected finding though was all children's accuracy – regardless of status – decreased during training and did not improve between baseline and follow-up. On pausing durations, children with dyslexia (singular and comorbid) took significantly longer to start writing alphabet letters than typically developing children. The visuomotor properties of novel characters also affected pausing behaviour during formation (intra-character pause durations). Across all analyses, children made shorter pauses within characters when copying b-type characters than d-type characters and two-stroke characters than three-stroke characters. Taken together, these findings

suggest that children – regardless of disorder status – were influenced by the visuomotor properties of characters prior to learning their motor programmes.

Most importantly, though, interactions revealed children with dyslexia-only and DCD-only paused for longer during formation after training. This impairment was probed further by examining performance during training. Analysis of the decrease in intra-character pausing durations (training values) revealed that the groups did not significantly differ from controls in the magnitude of the decrease of pausing durations during training. This suggests that impairments found after training were related to poor consolidation of motor programmes. Correlations revealed that intra-character pausing durations at follow-up were related to perceptuomotor ability and executive functions amongst children with dyslexia and with perceptuomotor ability amongst children with DCD. In what follows, these key findings are discussed in relation to the current literature. Relevant limiting conditions and implications are also discussed.

7.4.1. Legibility When Learning Novel Characters

Novel character formation was indexed using a binary accuracy scoring criteria in this study. Although it was not the primary outcome measure, it did generate some interesting and somewhat counterintuitive findings. Specifically, accuracy was found to decrease (nonsignificantly) during training and it did not improve between baseline and follow-up. Intuitively, one would expect accuracy to increase as children learn the motor programmes of the characters. It is important to consider explanations for these findings prior to discussing the findings in relation to pausing behaviours. A preliminary explanation for the decrease in accuracy during training was that children had become fatigued due to the length of the task and the level of concentration required to complete the task. It is likely that fatigue was psychological

rather than physiological because we did not observe any increase in pausing durations during training which could have indicated rest stops. It is likely then that psychological fatigue explains this decrease in accuracy during training, but it is unlikely that is played a role in the lack of improvement in accuracy between baseline and follow-up.

Finding no improvement in accuracy of forming novel characters between baseline and follow-up appears somewhat counter-intuitive, yet, the current findings are consistent with studies of early handwriting development. According to Graham et al. (1998), at the beginning of instruction, although children's handwriting fluency improves year on year, their handwriting legibility does not begin to improve until after fourth-grade (Year 5 in the UK). It appears then, that although handwriting fluency improves very rapidly, legibility improves at a much slower rate, which might explain the lack of improvement in accuracy but the decrease in pausing durations found here.

7.4.2. Motor Programme Learning Impairments in Dyslexia and DCD

As predicted, children with dyslexia-only and DCD-only made longer pauses and were therefore less fluent when forming characters after training. The position of these longer pauses – within characters – suggests children with dyslexia and/or DCD were spending longer monitoring formation (Chenu et al., 2014). Moreover, these children did not differ from controls in decreasing pausing duration during training. Taken together, these findings suggest that impairments in learning motor programmes were apparent in both dyslexia and DCD. These impairments did not appear to be related to problems in initially encoding motor information but were most likely related to impairments in consolidating motor programmes.

Although impairments in learning motor programmes were present in both dyslexia and DCD, it is possible that these impairments were related to different deficits in the disorders.

Correlations between pausing behaviours and factor score estimates from Study 2 revealed poorer fluency (relative to controls) at follow-up was related marginally to executive abilities in dyslexia. Since children with dyslexia had deficits in executive functions (see Study 2), it is possible that impairments in consolidating motor programmes were associated with executive function deficits in dyslexia. Correlations amongst children with DCD revealed poorer fluency (relative to controls) at follow-up was related to perceptuomotor ability only. Children with DCD had deficits in perceptuomotor skills (Study 2) it is therefore likely that difficulties in learning letter-like motor programmes were related to perceptuomotor deficits. It is important to stress, however, that these findings were correlational and, in some cases, marginally significant so the relations between deficits in executive functions and perceptuomotor skills and difficulties in acquiring new letters should be interpreted with caution.

In contrast to children with singular disorders, children with comorbid dyslexia and DCD made longer pauses than controls at baseline as well as follow-up. This was unexpected as previous studies have found children with comorbid dyslexia and DCD's profile of impairments to be similar to either or both singular disorders (see Studies 2 and 3). It is likely that this finding represents a statistical anomaly, especially given the small sample size and large variation within the comorbid group.

7.4.3. The Influence of Visuomotor Properties on Letter Formation

The literature suggests that children are sensitive to the visuomotor properties of letters prior to consolidating their motor programmes (e.g., Fischer, 2013; Thibon et al., 2018; Treiman & Kessler, 2011). It was reasoned that if children with dyslexia and/or DCD have difficulties in learning letter motor programmes then they may remain sensitive to the visuomotor properties of letters longer than typically developing children. Indeed, there is some evidence in the literature

to suggest that children with dyslexia and/or DCD are unduly influenced by the visual orientation and motor complexity of letters (e.g., Brooks et al., 2011; Prunty & Barnett, in press). Unexpectedly, however, the current findings suggest that all children in this study were sensitive to the visuomotor properties of novel characters, regardless of group membership. Thus, children with dyslexia and/or DCD were no more sensitive to visuomotor properties of characters than typically developing children.

In order to understand this unexpected finding, it is important to first consider how the current findings marry previous studies examining the influence of visuomotor properties on letter formation. In line with expectations, when writing highly familiar alphabet letters, children's fluency was not influenced by the visual orientation of the letter. This was expected because the current sample were, on average, older than the age where children are reported to be sensitive to the visual orientation of letters (Fischer, 2013; van Mier & Hulstijn, 1993). Children did, however, pause for longer – and were therefore less fluent – when copying letters with more strokes. This finding is consistent with Thibon et al. (2018) who also found dysfluency increased with an increasing number of strokes in children of a similar age. However, an issue with using alphabet letters is that it is hard to discern the impact of graphophonemic and visuomotor properties of the letter. Thus, a novel character learning paradigm – which controls potential phonological confounds – was used. This paradigm afforded the ability to examine whether children were sensitive to the visuomotor properties of orthographic characters when learning the motor programmes only.

When copying novel characters, children made shorter pauses when copying characters with a higher probabilistic b-type orientation which is consistent with how younger children write alphabet letters (Fischer, 2009; 2013; Treiman et al., 2014; Treiman & Kessler, 2011). This

suggests that, in the absence of consolidated motor programmes children rely on the statistical regularities of a letter's orientation (Treiman & Kessler, 2011; 2014). Children also made shorter pauses when writing novel characters with fewer strokes which corroborates findings by Thibon et al. (2018) who concluded that children were sensitive to the number of strokes in letters in the absence of a consolidated motor programme. Therefore, children who are unfamiliar with an orthographic character are sensitive to the visuomotor properties of the character.

As children became familiar with the new characters a change in pausing behaviours was also found. Before familiarisation – through training – children paused for less time prior to starting to write visuomotorically easier (b-type two-stroke) characters. After familiarisation, pre-writing pause durations no longer changed according to the character's visuomotor properties. Pre-writing pauses tap perception, retrieval, and planning processes (Chenu et al., 2014) and so this change in pausing behaviour before and after training could represent some consolidation of motor programmes. Motor programmes were unlikely to fully consolidate during training as this process takes years (Palmis et al., 2017) and children's intra-character pause durations continued to be influenced by the character's visuomotor properties after training indicating the motor programmes had not fully consolidated. Thus, the current findings are consistent with previous research regarding the influence of visuomotor properties of characters on letter formation processes. Finding consistency between the current study of older children learning novel characters and previous studies of younger children learning alphabet letters somewhat validates the methods employed for indexing visual orientation and motor complexity when learning new letters here. It is therefore unlikely that finding no difference in the effects of visuomotor properties of characters between children with or without dyslexia and/or DCD was attributable to the problems with the way visuomotor properties of letters were measured.

A potential explanation for the finding that children with dyslexia and/or DCD were no more sensitive to the visuomotor properties of the characters than typically developing children is that the latter had not yet consolidated motor programmes adequately for detectable differences. It follows that fully consolidating letter motor programmes takes several years of experience (e.g., Thibon et al., 2018) and the training in this study only provided eight trials of practice. Indeed, it is clear that complete consolidation was not possible as typically developing children remained sensitive to the visuomotor properties of characters at follow-up. Therefore, although this paradigm was able to detect group impairments in early consolidation; it was not sufficiently long enough for the letter programmes to consolidate in any group with respect to sensitivity to visuomotor properties of characters.

7.4.4. Translation from Novel Characters to Alphabet Letter Formation

The primary interest in the current study was to examine whether impairments in learning motor programmes could explain, in part, why children have letter formation difficulties. To examine learning it was important to control for experience of highly familiar alphabet letters – including knowledge of the letter's phonological form – by using novel orthographic characters. It is therefore important to consider how the current findings using novel characters translate to letter formation impairments in alphabet letters.

When copying novel characters, children's pausing behaviour suggested they were sensitive to the visuomotor properties of the character, as reported in studies with younger children writing alphabet letters (e.g., Thibon et al., 2018; Treiman & Kessler, 2011). Furthermore, moderate correlations were found between pausing durations of alphabet letters and novel characters. Taken together, this evidence suggests it is likely that pausing behaviours when forming novel characters were akin to pausing behaviours during early acquisition of alphabet

letters. Thus, the impairments in learning motor programmes of novel characters present in dyslexia and DCD may also be present when these children learn alphabet letters.

Learning motor programmes of alphabet letters is important for developing other aspects of letter knowledge (e.g., letter-sound knowledge). Studies with pre-literate children have demonstrated how training the motor component of letters via tracing led to greater gains in letter-sound knowledge, phonological awareness, and decoding ability, possibly by strengthening connections between phonological and orthographic representations (Bara et al., 2004; Hulme, 1979; Longcamp et al., 2005). This means that difficulties in learning to produce letters fluently early in development – as was found for children with dyslexia and DCD here – may result in, or enhance, difficulties in developing other aspects of letter knowledge. The consequences of difficulties in consolidating motor programmes amongst children with dyslexia and DCD on developing other aspects of letter knowledge requires further investigation.

7.4.5. Limiting Conditions

In considering how the current findings with novel characters translate to how children learn to form alphabet letters, it is important to consider some limitations in the current experimental paradigm. A rationale for using novel characters was that it was possible to control for confounds from existing phonological and graphemic knowledge of alphabet letters. However, when learning letter motor programmes, children learn phonological knowledge of the letter simultaneously. This phonological information likely bootstraps learning of motor programmes (e.g., Longcamp et al., 2005) and could protect against impairments or impede learning motor programmes (see Study 3).

Another limitation relates to the lack of complete consolidation of motor programmes in this study. Although the findings indicate some early consolidation of motor programmes was

taking place, all children remained sensitive to the visuomotor properties of the characters which suggests complete consolidation of the motor programmes had not been achieved. Therefore, training over eight consecutive blocks was not sufficient to achieve complete consolidation. For motor programmes to fully consolidate training would have to continue for several months and possibly years (Graham et al., 1998; Thibon et al., 2018). This means that the interpretation of the current findings is limited to early stages of learning (encoding and early consolidation) and claims cannot be made about later stages of learning letter motor programmes.

A final limitation to consider was potential interference of the surface texture on graphomotor behaviour during learning. The smoothness of the paper has been found to affect several graphomotor behaviours including letter formation, speed, and pausing (Alamargot & Morin, 2015; Chan & Lee, 2005; Wann & Nimmo-Smith, 1991). Low friction writing surfaces promote atypical graphomotor behaviours such as longer pausing durations, presumably by reducing the kinaesthetic feedback available to the child during writing (Alamargot & Morin, 2015; Wann & Nimmo-Smit, 1991). We minimised potential undue influence from low friction tablet surfaces by attaching a paper of the quality used in schools and offices (75 gsm) to the top of the tablet. When used in combination with the inking pen which features a biro tip, this setup closely approximates the friction children would typically be used to whilst writing (Alamargot & Morin, 2015). It is therefore unlikely that the experimental setup had any confounding effect on graphomotor behaviour in this study.

7.4.6. Conclusion

This study examined whether impairments in learning motor programmes could, in part, be responsible for letter formation difficulties in dyslexia and DCD. Impairments in learning motor programmes were probed by analysing pausing behaviours – a proxy for handwriting

fluency – whilst children learned novel orthographic characters varying in the visuomotor (orientation and motor) complexity. On the whole, children's pausing behaviours when learning novel characters were consistent with younger children learning alphabet letters. Moreover, impairments in early consolidation of motor programmes were apparent in both dyslexia and DCD. It is likely that impairments in learning motor programmes early in development, as demonstrated here, may directly and indirectly – via enhancing difficulties in learning other aspects of letter knowledge – lead to letter formation difficulties seen in dyslexia and DCD.

Chapter 8

General Discussion

Appropriate literacy development is not only important for educational attainment, but for social and economic outcomes too (Fisher & Twist, 2011). This thesis has focused on one aspect of literacy, writing development, amongst children who find learning to write most taxing. Foundational, or so-called transcription, skills of writing are spelling and handwriting, but these skills themselves are complex, taking several years to master (Berninger & Winn, 2006; Ehri, 2000; Graham et al., 1998). Children with dyslexia and DCD appear to have particular difficulties in mastering transcription skills (Berninger, 2008; Rosenblum et al., 2013). This thesis was devoted to understanding the nature of transcription difficulties in dyslexia and DCD.

To understand the nature of transcription difficulties in dyslexia and DCD we took a programmatic approach. The literature review highlighted that dyslexia and DCD share some overlap and are frequently comorbid (see Chapter 2). It was therefore important to better understand the relationship, if any, between the two disorders. Once the relationship between dyslexia and DCD had been clarified (Chapters 4 and 5), we were able to turn our attention to addressing the nature of transcription – particularly handwriting – difficulties in dyslexia and DCD (Chapters 6 and 7). In what follows, each of the studies of this thesis are briefly summarized. In summarizing each study, I discuss the key aims and findings as well as highlighting some specific implications. I then consider the findings in relation to the main aims of thesis, some overarching theoretical and practical implications and limiting conditions before discussing potential areas for further study.

8.1. Overview of The Main Findings

8.1.1. Prevalence of Literacy, Motor, and Comorbid Literacy and Motor Difficulties

In Study 1 (Chapter 4), the primary aim was to establish the prevalence of literacy, motor, and comorbid literacy and motor difficulties in a large unselected sample of children. The resulting sample ($N = 626$) of children aged 7.1 to 10.9 years old, revealed that

prevalence rates of literacy difficulties (7%) and motor difficulties (5%) closely

approximated previously reported prevalence rates of dyslexia-only and DCD-only

(Snowling & Hulme, 2015; Lingham et al., 2009). The rates of comorbid literacy and motor

difficulties (3%) were much smaller than previously reported for clinic and smaller

community-based samples (Cruddace & Riddell, 2006; Kaplan et al., 1998). Despite being

lower compared to other studies, the rate of comorbidity was much larger than expected by

chance based on the prevalence of singular disorders. Specifically, 17% of children with

markers of either literacy difficulties (dyslexia) or motor difficulties (DCD) had markers for

comorbid literacy and motor difficulty (comorbid dyslexia and DCD). These findings

highlight that comorbid literacy and motor difficulties (dyslexia and DCD) are common and

suggests the disorders could be related.

The high prevalence of comorbid dyslexia and DCD found in Study 1 suggested that

researchers and practitioners should at the very least screen for comorbid cases when

assessing for dyslexia or DCD. Potential relationships between frequently co-occurring

disorders are not consistent with single-deficit theories but are consistent with multifactorial

theories such as Pennington's (2006) multiple deficit model (MDM) which highlights that

comorbidity is to be expected. The relationship between dyslexia and DCD was not

addressed in this study and so the relationship between dyslexia and DCD was addressed in

Study 2 (Chapter 4).

8.1.2. Profiles of Markers in Dyslexia, DCD, and Comorbid Dyslexia and DCD

The focus of Study 2 (Chapter 5) was examining the relationship between dyslexia

and DCD. Firstly, we aimed to determine whether marker impairments in phonological,

executive, perceptuomotor, and literacy abilities were present in either or both of the

disorders. Secondly, we aimed to elucidate the nature of comorbid dyslexia and DCD. We

found that dyslexia was characterised by independent markers of phonological and literacy

impairments whereas DCD was characterised by independent markers of perceptuomotor impairments. Both dyslexia and DCD were characterised by shared markers of executive function impairments. Comorbid dyslexia and DCD was characterised by marker impairments in all abilities. The pattern and severity of impairments in comorbid dyslexia and DCD was an additive combination of impairments from singular dyslexia and/or DCD.

On a practical level, finding that children with comorbid dyslexia and DCD did not differ in the nature of impairments from children with singular dyslexia or DCD means the same assessment methods for either disorder can be used in combination to identify comorbid dyslexia and DCD. On a theoretical level, finding children with dyslexia and DCD to have deficits that were additive in nature was in line with the shared aetiology hypothesis of comorbidity and indicates comorbid dyslexia and DCD result from shared genetic factors. This again is consistent with the hypothesis from the MDM that comorbidity results from shared aetiologic (genetic and environmental) risk factors (Pennington, 2006).

Having elucidated the relationship and nature of dyslexia, DCD, and comorbid dyslexia and DCD we could turn our attention to the nature of transcription difficulties reported amongst children with these disorders. The findings from the previous study (Study 2) had already shed light onto the nature of spelling in these disorders, specifically showing that spelling impairments were present in dyslexia but not in DCD. We therefore focused our attention to understanding the nature of reported handwriting difficulties in dyslexia and DCD which remained unclear (Martlew, 1992; Rosenblum et al., 2013).

8.1.3. The Nature of Handwriting Difficulties in Dyslexia and DCD

In Study 3 (Chapter 6), the aim was to examine the nature of handwriting fluency and legibility difficulties reported in dyslexia and DCD (Martlew, 1992; Rosenblum et al., 2013). We found dissociable patterns of handwriting fluency and legibility impairments in dyslexia and DCD and these impairments were associated with deficits in handwriting related skills

identified in Study 2. When writing the alphabet, children with dyslexia had fluency

impairments whereas children with DCD did not. Among children with dyslexia's fluency

impairments stemmed from poor alphabet sequence knowledge, a literacy related deficit.

 On handwriting legibility, children with dyslexia had specific letter formation and word

spacing impairments. Letter formation impairments in this group were expected as this aspect

of handwriting taps alphabet knowledge (see Study 1). The small impairment in word spacing

in the dyslexia group was due to increased likelihood of segmenting word inappropriately at

prosodic boundaries, reflecting poor morphophonological and possibly lexical knowledge. In

comparison to children with dyslexia, children with DCD had difficulties in all aspects of

handwriting legibility (letter formation, letter spacing, word spacing, line alignment)

reflecting difficulties with motoric aspects of handwriting.

In children with dyslexia, phonological abilities were more strongly associated with

aspects of handwriting that were impaired and less so with unimpaired aspects of

handwriting. In children with DCD, perceptuomotor abilities were strongly associated with

aspects of handwriting that were impaired (legibility) and less so with unimpaired aspects of

handwriting (fluency). Taken together, profiles of handwriting fluency and legibility

impairments were different in dyslexia and DCD and predominantly aligned with the marker

impairments in dyslexia and DCD in Study 2 (Chapter 4). Thus, it is likely that handwriting

difficulties in dyslexia and DCD were related to impairments associated with the disorders

found in Study 2.

The findings in Study 3 highlight that handwriting reflects more than just motor

abilities but literacy-related processes, also (Abbott & Berninger, 1993). Furthermore,

handwriting impairments manifested in both dyslexia and DCD and so when assessing

handwriting difficulties, it is important for assessors to consider literacy as well as motor

deficits. Although handwriting impairments were largely dissociable between dyslexia and

DCD, children with either or both disorders had particular difficulties in letter formation as a specific feature of their handwriting. As such, examining possible bases for letter formation difficulties in dyslexia and DCD became the focus of the fourth study.

8.1.4. Impairments in Learning Novel Motor Programmes in Dyslexia and DCD

In this final study (Study 4, Chapter 7), impairments in learning letter motor programmes were examined as a possible contributing factor for later letter formation difficulties. The key finding in this study was children with dyslexia-only and DCD-only paused for longer and were therefore less fluent than typically developing children when forming newly-learned letters 1 to 3 days after training. The nature of this impairment suggested children were spending longer monitoring whilst forming the characters (Chenu et al., 2014), which was indicative of poorer consolidation of the motor programme. Poor consolidation of motor programmes is likely to lead to letter formation difficulties directly through degraded motor representations and possibly indirectly through mediating the development of other aspects of letter knowledge, such as letter-sound knowledge (Bara et al., 2004).

The presence of motor programme learning impairments found in this study raises some interesting implications. The presence of difficulties in forming letters fluently may be a useful early marker for identifying later handwriting difficulties. However, further (preferably longitudinal) work is necessary to establish the validity and utility of such a marker. Furthermore, the presence of motor programme learning impairments builds upon the findings regarding the nature of handwriting difficulties in dyslexia and DCD. The findings from Study 3, like those reported in the literature, indicated that handwriting difficulties in dyslexia and DCD are primarily associated with disorder-specific impairments (e.g., Sumner et al., 2013; Prunty et al., 2013). The findings from Study 4, however, suggest that handwriting difficulties in these disorders are also associated with more basic impairments in

learning. In this sense, handwriting difficulties are complex and multifaceted in nature, reflecting the multifactorial nature of dyslexia and DCD (Study 2).

8.1.5. Summary of the Main Findings

Having provided an overview of the main findings and some of their specific implications, what remains is to consider how these findings address the main aims of the thesis. Accordingly, there were two primary aims to this thesis. The first was to understand the relationship and comorbidity between dyslexia and DCD and the second was to understand the nature of spelling and handwriting difficulties in dyslexia and DCD. The current findings fulfil these aims.

In relation to the first aim, we found that dyslexia and DCD were characterised by multiple independent markers of phonological and literacy (dyslexia) or perceptuomotor (DCD) impairments. Yet, both disorders had shared markers of executive function impairments (Study 2, Chapter 5). In relation to comorbidity between dyslexia and DCD, we found both the high frequency of comorbidity (Study 1) and the additive profile of impairments from dyslexia and DCD found in comorbid dyslexia and DCD (Study 2) point to a shared aetiology between the disorders. Collectively, these findings go some way in explaining the relationship and comorbidity between dyslexia and DCD.

In relation to the second aim – to understand the nature of spelling handwriting difficulties in dyslexia and DCD – we found spelling impairments to be present in dyslexia but not DCD (Study 2). This resulted our attention being turned to handwriting. On handwriting, we found dissociable handwriting difficulties between dyslexia and DCD (Study 3). Different profiles of handwriting difficulties were associated with disorder-specific impairments. Specifically, impairments in alphabet writing, letter formation, and word spacing were associated with impairments in phonological and literacy skills in dyslexia whereas impairments in letter formation, letter spacing, word spacing, and line alignment

were associated with perceptuomotor skills in DCD (Studies 2 and 3). Exploring letter

formation impairments further, we found that impairments in learning motor programmes

were present in both dyslexia and DCD (Study 4). This means that handwriting difficulties

are likely to be associated with basic impairments in learning as well as disorder-specific

impairments. Thus, the current findings go some way in toward understanding the nature of

handwriting difficulties in dyslexia and DCD.

Finding different profiles of handwriting impairments in dyslexia and DCD which

were related to disorder specific impairments ultimately suggests that different aspects of

handwriting processing are impaired in the two disorders (e.g., van Galen, 1991). Studies

investigating neural correlates of handwriting processes corroborate this conclusion by

identifying separate cortical areas related to specific transcription processes (Purcell,

Turkeltaub, Eden, & Rapp, 2011). These cortical areas could be differentially impaired in

dyslexia and DCD. For example, the left fusiform gyrus (FG) and left inferior frontal gyrus

(IFG) are believed to be related to lexical and orthographic processing and are likely to be

areas of impairment in dyslexia (Purcell et al., 2011). Conversely, the left superior parietal

lobule (SPL) is an area associated with complex motor processing (Haaland, Elsinger, Mayer,

Durgerian, & Rao, 2004) and also implicated in writing (Purcell et al., 2011) and so it may be

an area impaired in DCD. A final cortical area of note is the left superior frontal gyrus (SFG),

or Exner's area, which is associated with allographic processing (Lubrano, Roux, &

Demonet, 2004). Allographic processing is believed to be reflected in letter formation to

some degree (Prunty & Barnett, in press) and so impairments in the left SFG may be present

in both dyslexia and DCD. Further work should test these hypothesised links between these

cortical areas and impaired handwriting in dyslexia and DCD. Nevertheless, having clarified

how the present findings meet the overall aims of the thesis, we now consider theoretical

implications to these findings.

8.2. Theoretical Implications

The findings from this thesis shed light on the relationship between dyslexia and DCD, and the comorbidity between the two disorders, as well as the transcription difficulties which manifest in these disorders. These findings contribute significantly to our burgeoning knowledge of the multifactorial nature of neurodevelopmental disorders and to theoretical models of writing.

8.2.1. Understanding the Nature of Neurodevelopmental Disorders

The current findings best fit with predictions from multifactorial (e.g., Bishop, 2006; Pennington, 2006) rather than single deficit accounts (see Chapter 2) of dyslexia and DCD on several fronts. A key differentiation between single and multiple deficit accounts is that the former predicts that only one deficit would be associated with a disorder whilst the latter account suggests multiple deficits would be present. Furthermore, the latter account also predicts that some deficits will be independent, and others will be shared (Bishop, 2006; Pennington, 2006). In line with the multifactorial account, multiple deficits were found in dyslexia and DCD. Some of these deficits were present only in dyslexia (e.g., phonological) or DCD (e.g., perceptuomotor) whilst others were shared (e.g., executive function) between the disorders.

Another point of corroboration between the current findings and multifactorial view is in explaining comorbidity. Single deficit theories do not account for comorbidity between disorders. For example, the internal modelling deficit of DCD (Wilson et al., 2004) does not explain why children with DCD frequently have comorbid dyslexia. Yet, the multifactorial account explicitly predicts this comorbidity. Pennington (2006) suggests that comorbidity is to be expected and results from shared aetiological risk factors. In line with these predictions, there was a high incidence of comorbidity between dyslexia and DCD. Moreover, the shared deficit in executive function skills points to at least one shared cognitive risk factor between

the two disorders. The additive nature of deficits in children with comorbid dyslexia and DCD further implies a shared genetic aetiology between the two disorders, also in line with the multifactorial account (Bishop, 2006; Pennington, 2006). It is clear then, that dyslexia and DCD are best considered within a multifactorial view.

Finding that dyslexia is best considered within a multifactorial view is not new. Previous studies have demonstrated the multifactorial nature of dyslexia, ADHD, and dyscalculia (Gooch et al., 2011; Moll et al., 2016; Pennington et al., 2011) and these findings are in line with current thinking about the nature of neurodevelopmental disorders more generally (Bishop, 2006; Snowling & Hulme, 2015; Thapar & Rutter, 2015). However, what is novel is considering DCD in relation to dyslexia as multifactorial. DCD is still often considered in single deficit terms, where one, two, or three deficits may be individually causally related to DCD (e.g., Wilson et al., 2017). However, the findings here suggest that deficits related to DCD and its comorbid disorders are best considered in a multifactorial manner. In this view, deficits such as the internal modelling deficit (see Chapter 2) are still related to DCD but are considered as a risk factor that acts probabilistically with other cognitive risk factors. To illustrate this argument, we consider how the findings from the current study fit within a simplified but extended version of the MDM (Pennington, 2006).

It was beyond the scope of this thesis to consider every level of analysis of the MDM (e.g., neural), however, some evidence can be considered at several levels of the model. At the highest (aetiological) level, genetic risk and protective factors of dyslexia and DCD act probabilistically. Some risk factors will be dyslexia- or DCD-specific, whilst others will be shared. These risk factors influence the development of neural systems, which in turn, influence the development of cognitive processes. Cognitive processes develop interactively where the atypical development of one set of processes will impact the development of other sets of processes. Deficits in cognitive processes – possibly such as phonological processing

– will increase the risk of dyslexia, whilst deficits in other cognitive processes – possibly

visuospatial processing – will increase the risk of DCD. Shared deficits – such as executive

functions – may also increase the risk of either one or both disorders. No one deficit is

sufficient or necessary for a disorder. These deficits or risk factors act probabilistically to

affect literacy and/or motor development and increase the risk of the impairment(s) reaching

a diagnostic threshold. Although the relationships between these markers and their respective

disorders was not tested, it is likely that some of these deficits will be causally related to

disorder (e.g., phonological deficits are causally related to dyslexia; Hulme et al., 2012)

whereas other deficits will compound difficulties increasing the risk of reaching diagnostic

threshold. For example, executive function deficits may compound phonological deficits in

dyslexia (Gathercole et al., 2016).

The MDM does not explicitly account for impairments in specific behaviours, such as

handwriting. Nevertheless, the findings of multifaceted handwriting difficulties in Studies 3

and 4 (Chapters 6 and 7, respectively) likely reflect the multifactorial nature of the dyslexia

and DCD. The dissociation of handwriting impairments likely reflects predominantly

different combinations of markers of dyslexia and DCD which were also skills related to

handwriting (Study 3). For example, handwriting difficulties amongst children with dyslexia,

in the main, were related to phonological and literacy related deficits, whereas handwriting

difficulties in DCD, in the main, were related to perceptuomotor deficits. In addition to these

deficits, handwriting difficulties – namely letter formation – likely also stemmed from a more

basic impairment in learning motor programmes early in development in both dyslexia and

DCD (Study 4). Therefore, findings from this thesis support the multifactorial view of

neurodevelopmental disorders and specifically expand Pennington's (2006) MDM to

consider dyslexia and DCD. Moreover, the findings from studies of handwriting difficulties

(Studies 3 and 4) further expand the MDM to consider how the multifactorial nature of disorders manifest in specific handwriting impairments.

8.2.2. Implications for Models of Writing

Spelling and handwriting skills – under the umbrella term of transcription skills – assume a pivotal position in models of writing development (Berninger & Swanson, 1994; Berninger & Winn, 2006). Transcription skills are considered foundational for writing where spelling and handwriting must become fluent or automatized to free up resources for higher level processes (Bourdin & Fayol, 1996; McCutcheon, 2011). Despite their prominent role, models of writing development often lump together spelling and handwriting and do not elaborate on their component processes. This lack of specificity presumably exists because most models of writing are concerned with mapping higher level processes (Hayes, 2012). Those models are useful for considering how impairments in transcription processes affect higher level writing processes (e.g., Connelly & Dockrell, 2016; Hayes & Berninger, 2014). However, the focus here was on how deficits relating to dyslexia and DCD manifest in transcription skills. The current findings point to some very specific impairments. For example, children with dyslexia have some impairments in specific aspects of handwriting legibility (letter formation and word spacing) but no impairment in others (letter spacing and line alignment). This dissection suggests that different processes are involved in handwriting production and a developmental model should account for this. Unfortunately, no model of writing development elaborates on spelling and handwriting processes or the interaction between the two. The model that gets closest to fitting this brief is van Galen's (1991) psychomotor model of skilled writing (c.f., Connelly, Dockrell, & Barnett, 2011).

8.2.2.1. The psychomotor model of writing. Whilst van Galen's (1991) psychomotor model of writing was not explicitly tested in this thesis, it was used as a framework for conceptualising how handwriting related skills contribute to handwriting

production (see Study 3). A test of the psychomotor model would be to consider whether the findings from the studies presented in this thesis match predictions generated by the model.

According to the psychomotor model, spelling processes cascade down into motor related processes, all with the support of buffers. A simple prediction from this model would be that impairments in spelling processing without motor processing impairments would result in handwriting difficulties directly reflecting spelling impairments. In contrast, impairments in motor but not spelling processes would result in handwriting difficulties reflecting motor impairments. The findings from Study 3, provide a natural test of this prediction because children with dyslexia had spelling related impairments but spared perceptuomotor skills whereas children with DCD had perceptuomotor related impairments but spared spelling skills. Whilst we were unable to test whether spelling or motor impairments directly predicted handwriting difficulties in these groups, we did find handwriting difficulties in children with dyslexia were related to impairments affecting spelling processes whereas handwriting difficulties in children with DCD were related impairments affecting motor processes. These findings are in line with predictions generated from van Galen's (1991) model.

The current evidence therefore is broadly consistent with cascading structure of van Galen's model, but the model should elaborate further on the relationship between spelling and motor processing. In particular, it is not clear how word spacing impairments found amongst children with dyslexia would be explained using the psychomotor model. Word spacing impairments reflected possible morphophonological deficits. Such language-based deficits, presumably in the spelling module, challenge the view of the model that words are activated as linear sequences of letters and instead suggests they are activated as functional linguistic units as highlighted by Kandel and colleagues (Kandel et al. 2009; Kandel et al., 2011; Kandel et al., 2012).

8.2.2.2. The limited capacity view of writing. The view that writing processes exert a cognitive load in a limited capacity system is a common one amongst writing researchers (Olive, 2014). In models of developing writing, the limited capacity view emphasises that children must automatize transcription processes in order to free up resources for higher level processes (Bourdin & Fayol, 1994; McCutcheon, 2011; Berninger et al., 2002). This theory is particularly relevant here as some argue that impaired/unautomatized spelling and/or motor related process in dyslexia and/or DCD place a high cognitive cost on the limited capacity system which limits the resources available for other processes, impacting on higher-level operations (McCutcheon, 2000; Sumner et al., 2016; Prunty et al., 2014). Whilst higher level writing processes were not examined in this thesis, the limited capacity theory would also assume that spelling and motor processes would compete for the same pool of limited resources in handwriting. Some consideration should therefore be made as to whether this theory can explain some of the current findings.

This thesis presents some mixed findings in relation to the limited capacity view. Broadly consistent with the expectations of limited capacity view, there were moderate to large correlations between handwriting and executive functions in the dyslexia and DCD groups but not in the typically developing group found in Study 3 (Chapter 6). This pattern of association could be interpreted as suggesting that children who were typically developing had unimpaired/automatized handwriting processes meaning more executive function resources were available for other processes. On the other hand, children with dyslexia and/or DCD had impaired handwriting processes which constrained the executive function resources available for other processes.

Due to its broad nature, the limited capacity view is hard to falsify (Torrance & Galbraith, 2006). However, some findings from Study 3 contradict predictions generated by the limited capacity view in relation to handwriting. The limited capacity view would predict

that impaired spelling and/or motor processes would heavily tax the available resources in all instances, leading to poorer handwriting and fluency. This means that impairments in motor processes should always constrain handwriting legibility and fluency in children with dyslexia and DCD. However, we found children with DCD did not have handwriting fluency impairments on an alphabet writing task despite having poor fine motor fluency (Study 2) and poor automatisation of motor processes at the level of letter legibility. Therefore, in this instance motor impairments did not constrain handwriting fluency which contrasts with the predictions of the limited capacity view.

Alternatively, an interference account, which suggests the output of one process interferes with another process (Torrance & Galbraith, 2006) could explain performance of both dyslexia and DCD on the alphabet writing task. The alphabet writing task relies heavily on alphabet sequence knowledge (Pontart et al., 2014). In the case of dyslexia, children paused for longer and made more sequencing type errors. According to the interference perspective, delays in retrieving alphabet sequence knowledge interfered with the motor production of the letters. In the case of DCD, children were not impaired, despite having motor processing impairments, because there was no interference from the retrieval of alphabet sequence knowledge. Although it was not an aim of this thesis to test the limited capacity or interference views of writing, these mixed findings highlight a potentially fruitful area for testing these explanations by comparing performance across disorders and tasks which place constraints on different processes of writing.

8.3. Implications for Practitioners and Researchers

Having discussed some key theoretical implications, it is now important to turn to consider some more practical applications of this work. The current findings have applications for researchers as well as practitioners concerned with classifying and ameliorating difficulties associated with dyslexia, DCD, and handwriting. Specific

implications in relation to assessment were highlighted when summarising findings of the studies earlier. Here, I elaborate on these implications to discuss them more generally. I also discuss some general implications in relation to intervention also.

8.3.1. Considerations for Assessment

There are two main implications for assessment that can be drawn from the current findings. The first is in relation to the assessment of dyslexia and DCD. The findings from Study 1 highlight that comorbidity between dyslexia and DCD is high. In this sample 17% of children who had literacy difficulties (dyslexia) or motor difficulties (DCD) had comorbid literacy and motor difficulties (dyslexia and DCD). Given the high likelihood for comorbidity it is important for researchers and practitioners to screen for additional disorders. The perils of not doing so are highlighted in the literature by a high degree of overlap in impairments which has subsequently been attributed to the presence of comorbid cases in the samples (see Study 2).

The nature of comorbid dyslexia and DCD is additive, meaning children with comorbid dyslexia and DCD have similar profiles of impairments to children with dyslexia-only and DCD-only (Study 2). An implication of this finding is that existing tests used to assess dyslexia and DCD can be used in concert to identify children with comorbid dyslexia and DCD, rather than having to develop specific tests to identify children with a comorbid disorder. The interested reader is directed to Snowling and Hulme (2015) who provide an overview of assessments for measuring multiple domains of math, reading, and writing ability which would capture frequently comorbid disorders affecting these abilities.

The findings in this thesis also relate to considerations regarding the assessment of handwriting difficulties amongst children with dyslexia and DCD. In the first instance, the findings highlight that it is important for handwriting ability to be assessed amongst children being tested for dyslexia and/or DCD. Whilst handwriting difficulties are considered

characteristic of DCD and such difficulties are often included in diagnostic criteria (e.g., APA, 2013) they are not considered diagnostic in dyslexia and are therefore not tested in dyslexia assessments. Yet, the findings in this thesis demonstrate that children with dyslexia have handwriting difficulties and therefore such difficulties should be investigated at assessment. It is not my position that handwriting difficulties should be included in diagnostic criteria for dyslexia, rather that handwriting assessments should be completed with children who are assessed for dyslexia with the view providing remedial assistance with handwriting difficulties if they are present.

When assessing handwriting in dyslexia and DCD, careful consideration is warranted in choosing the assessments. Assessments should be selected to capture the child's handwriting fluency and legibility abilities without tapping non-handwriting skills to a large degree. This point is highlighted by considering the findings when using a common alphabet writing task where performance appeared to be better explained by alphabet sequence knowledge rather than graphomotor fluency. A way of ensuring the validity of handwriting assessment protocols would be to administer multiple handwriting assessments.

8.3.2. Considerations for Intervention

The current findings also have implications for intervention. Practitioners who assess and identify comorbid dyslexia and DCD should consider and adjust for the comorbid disorder when ameliorating impairments relating to the core disorder. For example, when planning an intervention to improve reading and spelling impairments, practitioners should consider whether the activities such as writing may be particularly taxing for the child if they have comorbid dyslexia and DCD. Along the same lines, researchers should examine the efficacy of combining remedial programmes that target literacy and motor skills to improve outcomes for children with comorbid dyslexia and DCD.

With regards to ameliorating handwriting difficulties specifically, the current findings indicate that handwriting difficulties among children with dyslexia were specific and, in the main, reflected deficits in literacy-related abilities. The present results suggest that interventions concerning handwriting difficulties amongst children with dyslexia should combine reading and spelling activities with specific handwriting work focusing on letter formation and word spacing. Children with DCD, on the other hand, would likely benefit from more generalised handwriting work focusing on the motoric aspects of legibility in combination with a general motor-based intervention. Children with dyslexia and/or DCD also have difficulties in learning motor programmes of letters and so early interventions in particular would be useful in targeting this impairment. Further work is needed to rigorously examine the efficacy of providing targeted handwriting interventions to these groups.

8.4. Limiting Conditions

Specific limiting conditions have been noted within the chapters of this thesis. There are however some general limiting conditions across multiple studies which warrant some further discussion. Primary amongst these is the method used for assessing motor ability. Using a condensed battery of motor measures where time did not permit the use of a full battery has been used with some success in other studies (e.g., Lingham et al., 2009). A concern with this approach, however, is with issues relating to the use of standardised motor measures, including their reliability (Crawford et al., 2001; Venetsanou, 2011). Problems with reliability were apparent here also. Principle among motor measures with low reliability was the index of gross motor skill, Bag Throw. Indeed, this measure had to be removed from the CFA in Study 2 due to its poor fit. A reason for this poor fit could have been the large amount of measurement error. This means, though, that there were no reliable measures of gross motor skills in this series of studies. A solution for the future would be to include multiple measures of the same aspect of motor skills (e.g., fine motor, gross motor, etc.).

A primary aim of this thesis was to examine the frequent comorbidity between dyslexia and DCD. In addition, both dyslexia and DCD are frequently comorbid with ADHD (Kaplan et al., 1998) and transcription difficulties are also found in ADHD (Adi-Japha et al., 2007). Therefore, it would have been appropriate to include ADHD samples in the studies of this thesis. Whilst this would have been insightful, it was not feasible due to additional time required for recruiting ADHD samples with and without comorbid dyslexia and/or DCD.

A related issue is whether comorbid attentional difficulties were controlled for in the sample. To reduce the risk of comorbid ADHD in the sample, children were not recruited into the main sample (Phase 2 onwards) if they had received a diagnosis of ADHD. To ensure there was no undue influence of comorbid ADHD between children with dyslexia and/or DCD, or in typically developing children, groups were compared on a measure of sustained attention (the Score! subtest from the TEA-Ch; Manly et al., 1998), a cognitive marker impairment ADHD. This comparison revealed that the groups did not differ significantly on sustained attention ability, which suggests it was unlikely that comorbid ADHD influenced performance at the group level.

This thesis has focused on the relationship between children with clinical levels of impairments in literacy and/or motor skills and the effects of these impairments on transcription skills. To do so, we classified children as having dyslexia, DCD, or comorbid dyslexia and DCD by applying arbitrary cut-offs from continuous distributions. Indeed, Pennington (2006) acknowledges that applying cut-offs for disorders are somewhat arbitrary by stating, "The liability distribution for a given disease is often continuous and quantitative, rather than being discrete and categorical, so that the threshold for having the disorder is somewhat arbitrary." (Pennington, 2006, p. 404). By applying these cut-offs, we dichotomize variables with continuous distributions. It was deemed necessary to dichotomize variables with continuous distributions in this thesis because the focus was on children with clinical

levels of literacy and/or motor impairments and to identify similarities and differences between them. However, it is important to consider some of the implications in doing so. Such implications include loss of information of individual differences, reduction in power, increased risk of type 1 errors, and reduction in reliability (MacCallum, Zhang, Preacher, & Rucker, 2002).

In considering the relationship between spelling and motor processes relating to handwriting a unidirectional relationship was assumed where spelling processes affect lower motor processes. This direction was assumed based on the unidirectionality of the psychomotor model of writing (van Galen, 1991) where spelling processes cascade into motor processes but there is no reciprocal relationship between motor and spelling processes. This view was also somewhat validated by finding that impaired spelling processing impacted on motor processes in children with dyslexia yet, impaired motor processes did not impact spelling processing amongst children with DCD. However, studies with young children have demonstrated that training the motor aspects of letters leads to gains in early spelling abilities (e.g., Bara et al., 2004; Longcamp et al., 2005), suggesting a bidirectional relationship between motor and spelling processes, particularly in early writing. Further work should probe the potential reciprocal relationship between spelling and motor processes in early writing development.

8.5. Future Research

The current findings have produced several avenues for further investigation. Here, I limit my discussion of future research to two avenues. An interesting avenue to pursue and one that addresses the reciprocal relationship between motor and spelling processes would be to further investigate the relationship between learning letters' phonological versus motor form. The final study of this thesis used a novel orthographic character training paradigm to

elucidate impairments in learning motor programmes in children with dyslexia and DCD.

Yet, when learning to produce letters, children also learn the phonological form of the letter.

Learning the phonological form of letters will also impact on how well children are able to

learn to write them. The impact of learning either a letter's phonological form or its motor

programme, versus learning both forms on letter writing ability remains an unanswered but

interesting question. Particularly, in the cases of dyslexia and DCD, where children with

dyslexia have problems with acquiring letter-sound knowledge whereas children with DCD

presumably do not. Such a question could be addressed by including an additional

phonological training condition to the paradigm used in Study 4.

Addressing the relationship between language, motor skills, and handwriting further,

recent research suggests a link between oral-motor skills used in speech and the development

of phonological and reading ability. Specifically, oral-motor skills used in speech are related

to the development of phonological representations which, in turn, influence the development

of reading (van den Brunt et al., 2018, 2017). Such a link would imply that oral-motor skills

of speech are also likely to be indirectly linked to spelling and handwriting ability. However,

measuring oral-motor skills of speech production is difficult and has poor reliability (van den

Brunt, 2018). Therefore, future research should address the link between oral-motor skills of

speech production and transcription skills by (a) developing appropriate measures of oral-

motor skills and (b) examining the relationships between oral-motor skills and spelling and

handwriting ability, particularly in relation to children with literacy and motor difficulties.

The second avenue for further investigation would be to establish more concrete

evidence of a relationship between deficits in handwriting related skills and handwriting

difficulties in dyslexia and DCD. The correlational analyses between handwriting related

skills and handwriting reported here provided a good first step into examining these complex

relationships and support the behavioural profiles found on handwriting measures. However,

the interpretation of the correlational analyses is limited. Unfortunately, the current sample size of the dyslexia, DCD, and comorbid dyslexia and DCD groups did not permit the use of more powerful analyses to test the relationship between handwriting related skills and handwriting ability in dyslexia and DCD. Therefore, future work should build on the earlier work by Berninger and colleagues (Abbott & Berninger, 1993; Berninger et al., 1992; Berninger et al., 1994) to examine the key handwriting related skills of handwriting development and their impairments in children at risk of handwriting difficulties (e.g., dyslexia and DCD) in a large sample. Given the developmental nature of dyslexia and DCD, a longitudinal approach should be taken whereby the contribution of related skills in handwriting development should be assessed over time.

8.6. Conclusion

This thesis was devoted to understanding writing difficulties in dyslexia and DCD. To do so, a programmatic approach was undertaken, first to better understand the nature of the disorders themselves and second to understand the nature of handwriting difficulties apparent in these disorders. The studies reported in this thesis indicated that dyslexia and DCD have independent and shared impairments and are frequently comorbid with one another. The patterns of these impairments and comorbidity highlight the multifactorial nature of the disorders. The multifactorial nature of dyslexia and DCD was also apparent in their multifaceted handwriting difficulties. Handwriting difficulties in dyslexia and DCD manifested as dissociable impairments which reflected the nature of the specific disorder. In addition to these disorder-specific impairments, handwriting – specifically letter formation – difficulties also reflected impairments in early acquisition of handwriting related motor knowledge. Therefore, these investigations exploring the low-level aspects of transcription skills in dyslexia and DCD highlight the complex nature of writing difficulties. In doing so, this thesis has offered the first tentative steps in developing a rich understanding of the nature

of writing difficulties in dyslexia and DCD. With a better understanding of the nature of

difficulties, it is hoped we can develop effective interventions to ameliorate writing

difficulties in dyslexia and DCD.

References

Abbott, R. D., & Berninger, V. W. (1993). Structural equation modeling of relationships among developmental skills and writing skills in primary- and intermediate-grade writers. *Journal of Educational Psychology*, *85*(3), 478–508. doi: 10.1037//0022-0663.85.3.478

Accardo, A. P., Genna, M., & Borean, M. (2013). Development, maturation and learning influence on handwriting kinematics. *Human Movement Science*, *32*(1), 136–146. doi: 10.1016/j.humov.2012.10.004

Adams, I. L. J., Lust, J. M., Wilson, P., & Steenbergen, B. (2014). Compromised motor control in children with DCD: A deficit in the internal model?-A systematic review. *Neuroscience and Biobehavioral Reviews*, *47*, 225–244. doi: 10.1016/j.neubiorev.2014.08.011

Adi-Japha, E., Landau, Y. E., Frenkel, L., Teicher, M., Gross-tsur, V., & Shalev, R. S. (2007). ADHD and dysgraphia: Underlying mechanisms. *Cortex*, *43*(6), 700–709. doi: 10.1016/S0010-9452(08)70499-4

Alamargot, D., Caporossi, G., Chesnet, D., & Ros, C. (2011). What makes a skilled writer? Working memory and audience awareness during text composition. *Learning and Individual Differences*, *21*(5), 505–516. doi: 10.1016/j.lindif.2011.06.001

Alamargot, D., & Chanquoy, L. (2001). *Through the models of writing*. London, UK: Kluwer Academic Publishers. doi: 10.1007/978-94-010-0804-4

Alamargot, D., Chesnet, D., Dansac, C., & Ros, C. (2006). Eye and Pen: A new device for studying reading during writing. *Behavior Research Methods*, *38*(2), 287–299. doi: 10.3758/BF03192780

Alamargot, D., & Morin, M. F. (2015). Does handwriting on a tablet screen affect students' graphomotor execution? A comparison between grades two and nine. *Human Movement Science*, *44*(1), 32–41. doi: 10.1016/j.humov.2015.08.011

Alloway, T. P. (2007). Working memory, reading, and mathematical skills in children with developmental coordination disorder. *Journal of Experimental Child Psychology*, *96*, 20–36. doi: 10.1016/j.jecp.2006.07.002

Alloway, T. P. (2011). A comparison of working memory profiles in children with ADHD and DCD. *Child neuropsychology : a journal on normal and abnormal development in childhood and adolescence*, *17*(5), 483–94. doi: 10.1080/09297049.2011.553590

Alloway, T. P., & Temple, K. J. (2007). A Comparison of Working Memory Skills and Learning in Children with Developmental Coordination Disorder and Moderate

Learning Difficulties. *Applied Cognitive Psychology*, *21*, 473–487.

American Psychiatric Association. (1993). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Angold, A., Costello, E. J., & Erkanli, A. (1999). Comorbidity. *Journal of Child Psychology and Psychiatry*, *40*(1), 57–87.

Ans, B., & Carbonnel, S. (1998). A connectionist multiple-trace memory model for polysyllabic word reading. *Psychological Review*, *105*(4), 678–723.

Archibald, L. M. D., & Alloway, T. P. (2008). Comparing language profiles: Children with specific language impairment and developmental coordination disorder. *International Journal of Language & Communication Disorders*, *43*(2), 165–80. doi: 10.1080/13682820701422809

Bahr, R. H., Silliman, E. R., Berninger, V. W., & Dow, M. (2012). Linguistic pattern analysis of misspellings of typically developing writers in grades 1–9. *Journal of Speech, Language, and Hearing Research*, *55*(6), 1587–1599. doi: 10.1044/1092-4388(2012/10-0335)

Bairstow, P. J., & Laszlo, J. I. (1981). Kinaesthetic sensitivity to passive movements and its relationship to motor development and motor control. *Developmental Medicine and Child Neurology*, *23*, 606–616.

Bara, F., Gentaz, E., Colé, P., & Sprenger-Charolles, L. (2004). The visuo-haptic and haptic exploration of letters increases the kindergarten-children's understanding of the alphabetic principle. *Cognitive Development*, *19*, 433–449. doi: 10.1016/j.cogdev.2004.05.003

Bara, F., Morin, M. F., Alamargot, D., & Bosse, M. L. (2016). Learning different allographs through handwriting: The impact on letter knowledge and reading acquisition. *Learning and Individual Differences*, *45*, 88-94.

Barkley, R. A. (1997). Behavioral inhibition, sustained attention, and executive functions: "Constructing a unifying theory of ADHD". *Psychological Bulletin*, *121*(1), 65–94.

Barnett, A. L., Hill, E. L., Kirby, A., & Sugden, D. A. (2015). Adaptation and extension of the European Recommendations (EACD) on Developmental Coordination Disorder (DCD) for the UK context. *Physical & Occupational Therapy in Pediatrics*, *35*(2), 103–115.

Barnett, A., Henderson, S. E., Scheib, B., & Schulz, J. (2007). The detailed assessment of

speed of handwriting (DASH) [Assessment instrument]. London, UK: Pearson.

Barnett, A. L., Prunty, M., & Rosenblum, S. (2018). Development of the Handwriting Legibility Scale (HLS): A preliminary examination of reliability and validity. *Research in Developmental Disabilities*, *72*, 240–247. doi: 10.1016/j.ridd.2017.11.013

Beery, K. E., & Beery, N. A. (2010). Beery-Buktenica developmental test of visual-motor integration: Sixth edition (Beery VMI) [Assessment instrument]. Bloomington, US: Pearson.

Bellocchi, S., Muneaux, M., & Huau, A. (2017). Exploring the link between visual perception, visual–motor integration, and reading in normal developing and impaired children using DTVP-2. *Dyslexia*, *23*, 296–315. doi: 10.1002/dys.1561

Berninger, V. W. (1999). Coordinating transcription and text generation in working memory during composing: Automatic and constructive processes. *Learning Disability Quarterly*, *22*(2), 99–112.

Berninger, V. W., Abbott, R. D., Abbott, S. P., Graham, S., & Richards, T. (2002). Writing and reading: Connections between language by hand and language by eye. *Journal of Learning Disabilities*, *35*(1), 39–56. doi: 10.1177/002221940203500104

Berninger, V. W., Abbott, R. D., Jones, J., Wolf, B. J., Gould, L., Anderson-Youngstrom, M., … Apel, K. (2006). Early development of language by hand: Composing, reading, listening, and speaking connections; three letter-writing modes; and fast mapping in spelling. *Developmental Neuropsychology*, *29*(1), 61–92. doi: 10.1207/s15326942dn2901

Berninger, V. W., Abbott, R. D., Thomson, J. B., & Raskind, W. (2001). Language phenotype for reading and writing disability: A family approach. *Scientific Studies of Reading*, *5*(1), 59–106. doi: 10.1207/S1532799XSSR0501

Berninger, V. W., Abbott, R. D., Whitaker, D., Sylvester, L., & Nolen, S. B. (1995). Integrating low- and high-level skills in instructional protocols for writing disabilities. *Learning Disability Quarterly*, *18*(4), 293–309.

Berninger, V. W., & Amtmann, D. (2003). Preventing written expression disabilities through early and continuing assessment and intervention for handwriting and/or spelling problems: Research into practice. In H. L. Swanson, K. Harris, S. Graham (Eds.) *The handbook of learning disabilities* (pp.345-363).

Berninger, V. W., Cartwright, A., Yates, C., Swanson, H. L., & Abbott, R. D. (1994). Developmental skills related to writing and reading acquisition in the intermediate grades. *Reading and Writing*, *6*(2), 161–196. doi: 10.1007/BF01026911

Berninger, V. W., Mizokawa, D. T., & Bragg, R. (1991). Theory-based Diagnosis and Remediation of Writing Disabilities. *Journal of School Psychology*, *29*(1), 57–79. doi: 10.1016/0022-4405(91)90016-K

Berninger, V. W., Nielsen, K. H., Abbott, R. D., Wijsman, E., & Raskind, W. (2008). Writing problems in developmental dyslexia: under-recognized and under-treated. *Journal of school psychology*, *46*(1), 1–21. doi: 10.1016/j.jsp.2006.11.008

Berninger, V. W., & Rutberg, J. (1992). Relationship of finger function to beginning writing: Application to diagnosis of writing disabilities. *Developmental Medicine and Child Neurology*, *34*(3), 198–215.

Berninger, V. W., & Swanson, H. L. (1994). Modifying Hayes and Flower's model of skilled writing to explain beginning and developing writing. In E. C. Butterfield, & J. S. Carlson (Eds.), *Advances in cognition and educational practice*, 2, (pp. 57-81).

Berninger, V., & Winn, W. (2006). Implications of advancements in brain research and technology for writing development, writing instruction, and educational evolution. *Handbook of writing research,* (pp. 96-114).

Berninger, V. W., Yates, C., Cartwright, A., Rutberg, J., Remy, E., & Abbott, R. D. (1992). Lower-level developmental skills in beginning writing. *Reading and Writing*, *4*(3), 257–280. doi: 10.1007/BF01027151

Biotteau, M., Chaix, Y., & Albaret, J. (2015). Procedural learning and automatization process in children with developmental coordination disorder and/or developmental dyslexia. *Human Movement Science*, *43*, 78–89. doi: 10.1016/j.humov.2015.07.005

Bishop, D. V. M. (2006). Developmental cognitive genetics: How psychology can inform genetics and vice versa. *The Quarterly Journal of Experimental Psychology*, *59*(7), 1153–1168. doi: 10.1080/17470210500489372

Bishop, D. V. M. (2010). Which neurodevelopmental disorders get researched and why? *PLoS one*, *5*(11), e15112. doi: 10.1371/journal.pone.0015112

Blank, R., Smits-Engelsman, B., Polatajko, H., & Wilson, P. (2012). European Academy for Childhood Disability (EACD): Recommendations on the definition, diagnosis and intervention of developmental coordination disorder (long version). *Developmental medicine and child neurology*, *54*(1), 54–93. doi: 10.1111/j.1469-8749.2011.04171.x

Bonifacci, P. (2004). Children with low motor ability have lower visual-motor integration ability but unaffected perceptual skills. *Human Movement Science*, *23*(2), 157–168. doi: 10.1016/j.humov.2004.08.002

Bosse, M., Tainturier, M.-J., & Valdois, S. (2007). Developmental dyslexia: The visual

attention span deficit hypothesis. *Cognition*, *104*(2), 198–230. doi: 10.1016/j.cognition.2006.05.009

Bosse, M., & Valdois, S. (2009). Influence of the visual attention span on child reading performance: A cross-sectional study. *Journal of Re*, *32*(2), 230–253. doi: 10.1111/j.1467-9817.2008.01387.x

Both-de Vries, A. C., & Bus, A. G. (2008). Name Writing: A First Step to Phonetic Writing? Does the Name Have a Special Role in Understanding the Symbolic Function of Writing? *Literacy Teaching and Learning*, *12*(2), 37–55.

Bourassa, D. C., & Treiman, R. (2003). Spelling in children with dyslexia: Analyses from the Treiman-Bourassa early spelling test. *Scientific Studies of Reading*, *7*(4), 309–333. doi: 10.1207/s1532799xssr0704_1

Bourdin, B., & Fayol, M. (1994). Is written language production more difficult than oral language production? A working memory approach. *International Journal of Psychology*, *29*(5), 561620. doi: 10.1080/00207599408248175

Brooks, A. D., Berninger, V. W., & Abbott, R. D. (2011). Brooks, A. D., Berninger, V. W., & Abbott, R. D. (2011). Letter naming and letter writing reversals in children with dyslexia: Momentary inefficiency in the phonological and orthographic loops of working memory. *Developmental Neuropsychology*, *36*(7), 847–868. doi: 10.1080/87565641.2011.606401

Brown, T. A. (2015). *Confirmatory factor analysis for applied research*.

Byrne, B. (1998). The foundation of literacy: The child's acquisition of the alphabetic principle. Hove, East Sussex: Psychology Press.

Caravolas, M. (2004). Spelling development in alphabetic writing systems: A cross-linguistic perspective. *European Psychologist*, *9*(1), 3–14. doi: 10.1027/1016-9040.9.1.3

Caravolas, M. (2017). Growth of word and pseudoword reading efficiency in alphabetic orthographies: Impact of consistency. *Journal of Learning Disabilities*, *51*(5), 422–433. doi: 10.1177/0022219417718197

Caravolas, M. & Downing, C. R. (in prep). Spelling and Handwriting Legibility Test (SaLHT) [Assessment Instrument].

Caravolas, M., Hulme, C., & Snowling, M. J. (2001). The foundations of spelling ability: Evidence from a 3-year longitudinal study. *Journal of Memory and Language*, *45*(4), 751–774. doi: 10.1006/jmla.2000.2785

Caravolas, M., Lervåg, A., Defior, S., Seidlová Málková, G., & Hulme, C. (2013). Different patterns, but equivalent predictors, of growth in reading in consistent and inconsistent

orthographies. *Psychological science*, *24*(8), 1398–407. doi: 10.1177/0956797612473122

Caravolas, M., Lervåg, A., Mousikou, P., Efrim, C., Litavsky, M., Onochie-Quintanilla, E., … Hulme, C. (2012). Common patterns of prediction of literacy development in different alphabetic orthographies. *Psychological science*, *23*(6), 678–86. doi: 10.1177/0956797611434536

Caravolas, M., & Samara, A. (2015). Learning to read and spell words in different writing systems. In A. Pollatsek, & R. Treiman (Eds.), *The Oxford handbook of reading* (pp. 326-343).

Caravolas, M., & Volín, J. (2001). Phonological spelling errors among dyslexic children learning a transparent orthography: The case of Czech. *Dyslexia*, *7*(4), 229–245. doi: 10.1002/dys.206

Caravolas, M., Volín, J., & Hulme, C. (2005). Phoneme awareness is a key component of alphabetic literacy skills in consistent and inconsistent orthographies: Evidence from Czech and English children. *Journal of Experimental Child Psychology*, *92*(2), 107–139. doi: 10.1016/j.jecp.2005.04.003

Caron, C., & Rutter, M. (1991). Comorbidity in Child Psychopathology: Concepts , Issues and Research Strategies. *Journal of Child Psychology and Psychiatry*, *32*(7), 1063–1080.

Casco, C., Tressoldi, P. E., & Dellantonio, A. (1998). Visual selective attention and reading efficiency are related in children. *Cortex*, *34*(4), 531–546.

Castellanos, F. X., & Tannock, R. (2002). Neuroscience of attention-deficit/hyperactivity disorder: The search for endophenotypes. *Nature Reviews Neuroscience*, *3*(8), 617. doi: 10.1038/nrn896

Catts, H. W., Adlof, S. M., Hogan, T. P., & Weismer, S. E. (2005). Are Specific Language Impairment and Dyslexia Distinct Disorders? *Journal of Speech, Language, and Hearing Research*, *48*(6), 1378–1396. doi: 10.1044/1092-4388(2005/096)

Chenoweth, N. A., & Hayes, J. R. (2001). Fluency in writing: Generating text in L1 and L2. *Written Communication*, *18*(1), 80-98.

Chenu, F., Pellegrino, F., Jisa, H., & Fayol, M. (2014). Interword and intraword pause threshold in writing. *Frontiers in Psychology*, *5*(182), 1–7. doi: 10.3389/fpsyg.2014.00182

Cheung, G. W., & Rensvold, R. B. (2002). Evaluating goodness-of-fit indexes for testing measurement invariance. *Structural Equation Modeling: A Multidisciplinary Journal*,

9(2), 233–255. doi: 10.1207/S15328007SEM0902

Cirino, P. T., Ahmed, Y., Miciak, J., Taylor, W. P., Gerst, E. H., & Barnes, M. A. (2018). A Framework for Executive Function in the Late Elementary Years. *Neuropsychology*, *32*(2), 176–189. doi: 10.1037/neu0000427

Coleman, R., Piek, J. P., & Livesey, D. J. (2001). A longitudinal study of motor ability and kinaesthetic acuity in young children at risk of developmental coordination disorder. *Human Movement Science*, *20*, 95–110.

Collette, M. A. (1979). Dyslexia and classic pathogenic signs. *Perceptual and Motor Skills*, *48*, 1055–1062.

Coltheart, M. (2005) Modeling reading: The dual-route approach. In M. J. Snowling & C. Hulme (Eds.) *The science of reading: A handbook* (pp. 6-23).

Connelly, V., Campbell, S., & Maclean, M. (2006). Contribution of lower order skills to the written composition of college students with and without dyslexia. *Developmental Neuropsychology*, *29*(1), 37–41. doi: 10.1207/s15326942dn2901

Cooke, A. (2002). Case study: A virtual non-reader achieves a degree. *Dyslexia*, *8*(2), 102–15. doi: 10.1002/dys.217

Cornhill, H., & Case-Smith, J. (1996). Factors that relate to good and poor handwriting. *The American Journal of Occupational Therapy*, *50*(9), 732–739.

Correa, J., & Dockrell, J. E. (2007). Unconventional word segmentation in Brazilian children's early text production. *Reading and Writing*, *20*, 815–831. doi: 10.1007/s11145-006-9049-3

Crammond, D. J. (1997). Motor imagery: Never in your wildest dream. *Trends in Neurosciences*, *20*(2), 54–57.

Crawford, S. G., & Dewey, D. (2008). Co-occurring disorders: A possible key to visual perceptual deficits in children with developmental coordination disorder? *Human Movement Science*, *27*(1), 154–169. doi: 10.1016/j.humov.2007.09.002

Crawford, S. G., Wilson, B. N., & Dewey, D. M. (2001). Identifying developmental coordination disorder: Consistency between tests. *Physical & Occupational Therapy in Pediatrics*, *20*(2–3), 29–50. doi: 10.1300/J006v20n02

Cruddace, S. A., & Riddell, P. M. (2006). Attention processes in children with movement difficulties, reading difficulties or both. *Journal of Abnormal Child Psychology*, *34*(5), 675–683. doi: 10.1007/s10802-006-9053-8

Cutler, A., & Butterfield, S. (1990). Durational cues to word boundaries in clear speech. *Speech Communication*, *9*(5–6), 485–495.

Dandache, S., Wouters, J., & Ghesquière, P. (2014). Development of reading and phonological skills of children at family risk for dyslexia : A longitudinal analysis from kindergarten to sixth grade. *Dyslexia*, *20*, 305–329. doi: 10.1002/dys.1482

Danna, J., & Velay, J. (2015). Basic and supplementary sensory feedback in handwriting. *Frontiers in Psychology*, *6*(169), 1–11. doi: 10.3389/fpsyg.2015.00169

de Jong, C. G. W., Oosterlaan, J., & Sergeant, J. A. (2006). The role of double dissociation dtudies in the dearch for dandidate endophenotypes for the comorbidity of attention deficit hyperactivity disorder and reading disability. *International Journal of Disability, Development and Education*, *53*(March 2015), 177–193. doi: 10.1080/10349120600716158

Decker, S. L., Roberts, A. M., & Englund, J. A. (2013). Cognitive predictors of rapid picture naming. *Learning and Individual Differences*, *25*(June), 141–149. doi: 10.1016/j.lindif.2013.03.009

Delattre, M., Bonin, P., & Barry, C. (2006). Written spelling to dictation: Sound-to-spelling regularity affects both writing latencies and durations. *Journal of Experimental Psychology: Learning, memory, and cognition*, *32*(6), 1330–40. doi: 10.1037/0278-7393.32.6.1330

Dewey, D. M., Kaplan, B. J., Crawford, S. G., & Wilson, B. N. (2002). Developmental coordination disorder: Associated problems in attention, learning, and psychosocial adjustment. *Human Movement Science*, *21*(5–6), 905–918. doi: 10.1016/S0167-9457(02)00163-X

Diamond, A. (2013). Executive functions. *Annual review of psychology*, *64*, 135–168. doi: 10.1146/annurev-psych-113011-143750

Dirks, E., Spyer, G., van Lieshout, E. C. D. M., & de Sonneville, L. (2008). Prevalence of combined reading and arithmetic disabilities. *Journal of Learning Disabilities*, *41*(5), 460–473. doi: 10.1177/0022219408321128

Ehri, L. C. (1997). Learning to read and learning to spell are one and the same, almost. In C. A. Perfetti, l. Rieben, & M. Fayol (Eds.), *Learning to spell: Research, theory, and practice across languages* (pp. 237-268).

Fawcett, A. J., Nicolson, R. L., & Dean, P. (1996). Impaired performance of children with dyslexia on a range of cerebellar tasks. *Annals of Dyslexia*, *46*(1), 259–283.

Feder, K. P., & Majnemer, A. (2007). Handwriting development, competency, and intervention. *Developmental Medicine and Child Neurology*, *49*, 312–317.

Fischer, F. . W., Liberman, I. Y., & Shankweiler, D. (1978). Reading reversals and

developmental dyslexia: A further study. *Cortex*, *14*(4), 496–510. doi: 10.1016/S0010-9452(78)80025-2

Fisher, R. & Twist, L. (2011). *Evaluation of every child a writer: Report 1* (Report No. DFE-RR108(a). Retrieved from https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/182503/DFE-RR108A.pdf

Fischer, J. (2011). Mirror writing of digits and (capital) letters in the typically developing child. *Cortex*, *47*(6), 759–762. doi: 10.1016/j.cortex.2011.01.010

Fischer, J. (2013). Digit reversal in children's writing: A simple theory and its empirical validation. *Journal of Experimental Child Psychology*, *115*(2), 356–370. doi: 10.1016/j.jecp.2013.02.003

Fletcher, J. M., Shaywitz, S. E., Shankweiler, D. P., Katz, L., Liberman, I. Y., Stuebing, K. K., … Shaywitz, B. A. (1994). Cognitive profiles of reading disability: Comparisons of discrepancy and low achievement definitions. *Journal of Educational Psychology*, *86*(I), 6–23.

Flower, L., & Hayes, J. R. (1981). A cognitive process theory of writing. *College Composition and Communication*, *32*(4), 365–387.

Gardner, G. T. (1996). Test of Visual Perception Skills (TVPS) [Assessment instrument]. London, UK: Ann Arbour

Gathercole, S., & Pickering, S. (2001). Working memory test battery for children (WMTB-C) [Assessment instrument]. London, UK: Pearson.

Gathercole, S. E., Woolgar, F., Kievit, R. A., Astle, D., Manly, T., Holmes, J., & CALM Team. (2016). How common are WM deficits in children with difficulties in reading and mathematics? *Journal of Applied Research in Memory and Cognition*, *5*(4), 384–394. doi: 10.1016/j.jarmac.2016.07.013

Georgiou, G. K., Torppa, M., Manolitsis, G., Lyytinen, H., & Parrila, R. (2012). Longitudinal predictors of reading and spelling across languages varying in orthographic consistency. *Reading and Writing*, *25*(2), 321–346. doi: 10.1007/s11145-010-9271-x

Germanò, E., Gagliano, A., & Curatolo, P. (2010). Comorbidity of ADHD and Dyslexia. *Developmental Neuropsychology*, *35*(5), 475–493. doi: 10.1080/875656412010494748

Glascoe, F. P., & Byrne, K. E. (1993). The accuracy of three developmental screening tests. *Journal of Early Intervention*, *17*(4), 368–379. doi: 10.1177/105381519301700403

Glutting, J. J., Adams, W., & Sheslow, D. (2000). Wide Range Intelligence Test [Assessment instrument]. Wilmington, DE: Wide Range.

Gomez, A., & Sirigu, A. (2015). Developmental coordination disorder: Core sensori-motor deficits, neurobiology and etiology. *Neuropsychologia*, *79*, 272–287. doi: 10.1016/j.neuropsychologia.2015.09.032

Gooch, D., Hulme, C., Nash, H. M., & Snowling, M. J. (2014). Comorbidities in preschool children at family risk of dyslexia. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, *55*, 237–246. doi: 10.1111/jcpp.12139

Gooch, D., Snowling, M. J., & Hulme, C. (2011). Time perception, phonological skills and executive function in children with dyslexia and/or ADHD symptoms. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, *52*, 195–203. doi: 10.1111/j.1469-7610.2010.02312.x

Gosse, C., Carbonnelle, S., de Vleeschouwer, C., & Van Reybroeck, M. (2018). Specifying the graphic characteristics of words that influence children's handwriting. *Reading and Writing*, *31*(5), 1181–1207. doi: 10.1007/s11145-018-9834-9

Goswami, U. (2015a). Sensory theories of developmental dyslexia: Three challenges for research. *Nature Reviews Neuroscience*, *16*(1), 43–54. doi: 10.1038/nrn3836

Goswami, U. (2015b). Visual attention span deficits and assessing causality in developmental dyslexia. *Nature Reviews Neuroscience*, *16*(4), 225. doi: 10.1038/nrn3836-c2

Gough, P. B., & Tunmer, W. E. (1986). Decoding, reading, and reading disability. *Remedial and Special Education*, *7*(1), 6–10. doi: 10.1177/074193258600700104

Graham, S. (1986). The reliability , validity , and utility of three handwriting measurement procedures. *The Journal of Educational Research*, *79*(6), 373–380.

Graham, S., Berninger, V. W., Abbott, R. D., Abbott, S. P., & Whitaker, D. (1997). Role of mechanics in composing of elementary school students: A new methodological approach. *Journal of Educational Psychology*, *89*(1), 170–182. doi: 10.1037/0022-0663.89.1.170

Graham, S., Berninger, V. W., Weintraub, N., & Schafer, W. (1998). Development of handwriting speed and legibility in grades 1–9. *The Journal of Educational Research*, *92*(June), 42–52. doi: 10.1080/00220679809597574

Graham, S., & Harris, K. R. (2005). Improving the writing performance of young struggling writers: Theoretical and programmatic research from the center on accelerating student learning. *The Journal of Special Education*, *39*(1), 19–33.

Graham, S., Harris, K. R., Mason, L., Fink-Chorzempa, B., Moran, S., & Saddler, B. (2008). How do primary grade teachers teach handwriting? A national survey. *Reading and Writing*, *21*(1–2), 49–69. doi: 10.1007/s11145-007-9064-z

Graham, S., Mckeown, D., Kiuhara, S., & Harris, K. R. (2012). A meta-analysis of writing instruction for students in the elementary grades. *Journal of Educational Psychology*, *104*(4), 879–896. doi: 10.1037/a0029185

Graham, S., Struck, M., Santoro, J., & Berninger, V. W. (2006). Dimensions of good and poor handwriting legibility in first and second graders: Motor programes, visual-spatial arrangement, and letter formation parameter setting. *Developmental Neuropsychology*, *29*(1), 43–60. doi: 10.1207/ s15326942dn2901_4

Grice, J. W. (2001). Computing and evaluating factor scores. *Psychological Methods*, *6*(4), 430–450. doi: 10.1037//1082-989x.6.4.430

Halsband, U., & Lange, R. K. (2006). Motor learning in man: A review of functional and clinical studies. *Journal of Physiology-Paris*, *99*(4), 414–424. doi: 10.1016/j.jphysparis.2006.03.007

Hamstra-Bletz, L., & Blöte, A. W. (1993). A longitudinal study on dysgraphic handwriting in primary school. *Journal of Learning Disabilities, 26*(10), 689-699. doi: 10.1177/002221949302601007

Hawelka, S., & Wimmer, H. (2008). Visual target detection is not impaired in dyslexic readers. *Vision Research*, *48*(6), 850–852. doi: 10.1016/j.visres.2007.11.003

Hayes, J. R. (1996). A new framework for understanding cognition and affect in writing. In C. M. Levy & S. Randall (Eds.), *The science of writing: Theories, methods, individual differences, and applications* (pp. 1-27). Mahwah, NJ: Erlbaum.c

Hayes, J. R. (2012). Modeling and remodeling writing. *Written Communication*, *29*(3), 369–388. doi: 10.1177/0741088312451260

Hayes, J. R., & Berninger, V. W. (2014). Cognitive processes in writing: A framework. *Writing development in children with hearing loss, dyslexia, or oral language problems: Implications for assessment and instruction*, (pp. 3-15).

Hayes, J. R., & Chenoweth, N. A. (2006). Is working memory involved in the transcribing and editing of texts? *Written Communication*, *2*, 135–149. doi: 10.1177/0741088306286283

Hayes, J. R., & Flower, L. S. (1980). Identifying the organization of writing processes. In Gregg, L. W. & Steinberg, E. R. (Eds.), *Cognitive processes in writing* (pp. 3-30).

Henderson, S. E., Barnett, A. L., & Henderson, L. (1994). Visuospatial difficulties and clumsiness: On the interpretation of conjoined deficits. *Journal of Child Psychology and Psychiatry*, *35*(5), 961–969.

Henderson, S., & Sugden, D., (1992). Movement assessment battery for children (M-ABC)

[Assessment instrument]. London, UK: Pearson.

Henderson, S., Sugden, D., & Barnett, A. (2007). Movement assessment battery for children-2: Second edition (M-ABC 2) [Assessment instrument]. London, UK: Pearson.

Hill, E. L. (2001). Non-specific nature of specific language impairment: A review of the literature with regard to concomitant motor impairments. *International Journal of Language & Communication Disorders*, *36*(2), 149–171. doi: 10.1080/13682820010019874

Hooper, Stephen, R., Costa, L.-J., Mcbee, M., Anderson, K. L., Yerby, D. C., Knuth, S. B., & Childress, A. (2011). Concurrent and longitudinal neuropsychological contributors to written language expression in first and second grade students. *Reading and Writing*, *24*, 221–252. doi: 10.1007/s11145-010-9263-x

Houghton, G., & Zorzi, M. (2003). Normal and impaired spelling in a connectionist dual-route architecture. *Cognitive Neuropsychology*, *20*(2), 115–62. doi: 10.1080/02643290242000871

Hu, L., & Bentler, P. M. (1999). Cutoff criteria for fit indexes in covariance structure analysis: Conventional criteria versus new alternatives. *Structural Equation Modeling: A Multidisciplinary Journal*, *6*(1), 1–55. doi: 10.1080/10705519909540118

Huau, A., Velay, J., & Jover, M. (2015). Graphomotor skills in children with developmental coordination disorder (DCD): Handwriting and learning a new letter. *Human Movement Science*, *42*, 318–332.

Hulme, C. (1979). The interaction of visual and motor memory for graphic forms following tracing. *The Quarterly Journal of Experimental Psychology*, *31*(2), 249-261.

Hulme, C., Bowyer-Crane, C., Carroll, J. M., Duff, F. J., & Snowling, M. J. (2012). The Causal role of phoneme awareness and letter-sound knowledge in learning to read: Combining intervention studies with mediation analyses. *Psychological Science*, *23*(6), 572–577. doi: 10.1177/0956797611435921

Hulme, C., Nash, H. M., Gooch, D., Lervåg, A., & Snowling, M. J. (2015). The foundations of literacy development in children at familial risk of dyslexia. *Psychological Science*, *26*(12), 1877–86. doi: 10.1177/0956797615603702

Hulme, C., Smart, A., & Moran, G. (1982). Visual perceptual deficits in clumsy children. *Neuropsychologia*, *20*(4), 475–481.

Hulme, C., & Snowling, M. J. (2013). *Developmental disorders of language learning and cognition*. John Wiley & Sons.

Iversen, S., Berg, K., Ellertsen, B., & Tønnessen, F.-E. (2005). Motor coordination

difficulties in a municipality group and in a clinical sample of poor readers. *Dyslexia*, *11*(3), 217–231. doi: 10.1002/dys.297

Jeannerod, M. (2006). *Motor Cognition: What Actions Tell the Self* (1st ed). Oxford: Oxford University Press.

Jones, D., & Christensen, C. A. (1999). Relationship between automaticity in handwriting and students' ability to generate written text. *Journal of Educational Psychology*, *91*(1), 44–49.

Jover, M., Schmitz, C., Centelles, L., & Chabrol, B. (2010). Anticipatory postural adjustments in a bimanual load-lifting task in children with developmental coordination disorder. *Developmental Medicine and Child Neurology*, *52*(9), 850–855. doi: 10.1111/j.1469-8749.2009.03611.x

Juel, C., Griffith, P. L., & Gough, P. B. (1986). Acquisition of literacy: A longitudinal study of children in first and second grade. *Journal of Educational Psychology*, *78*(4), 243–255.

Kadesjö, B., & Gillberg, C. (1999). Developmental coordination disorder in Swedish 7-year-old children. *Journal of the American Academy of Child and Adolescent Psychiatry*, *38*(7), 820–8. doi: 10.1097/00004583-199907000-00011

Kandel, S., Hérault, L., Grosjacques, G., Lambert, E., & Fayol, M. (2009). Orthographic vs . phonologic syllables in handwriting production. *Cognition*, *110*(3), 440–444. doi: 10.1016/j.cognition.2008.12.001

Kandel, S., Lassus-sangosse, D., Grosjacques, G., & Perret, C. (2017). The impact of developmental dyslexia and dysgraphia on movement production during word writing. *Cognitive Neuropsychology*, *34*(3–4), 219–251. doi: 10.1080/02643294.2017.1389706

Kandel, S., Peereman, R., Grosjacques, G., & Fayol, M. (2011). For a psycholinguistic model of handwriting production: Testing the syllable-bigram controversy. *Journal of Experimental Psychology: Human Perception and Performance*, *37*(4), 1310–1322. doi: 10.1037/a0023094

Kandel, S., & Perret, C. (2015). How does the interaction between spelling and motor processes build up during writing acquisition? *Cognition*, *136*, 325–336. doi: 10.1016/j.cognition.2014.11.014

Kandel, S., Spinelli, E., Tremblay, A., Guerassimovitch, H., & Álvarez, C. J. (2012). Processing prefixes and suffixes in handwriting production. *Acta Psychologica*, *140*(3), 187–195. doi: 10.1016/j.actpsy.2012.04.005

Kaplan, B. J., Dewey, D. M., Crawford, S. G., & Wilson, B. N. (2001). The term comorbidity

is of questionable value in reference to developmental disorders: Data and theory. *Journal of learning disabilities*, *34*(6), 555–565.

Kaplan, B. J., Wilson, B. N., Dewey, D. M., & Crawford, S. G. (1998). DCD may not be a discrete disorder. *Human Movement Science*, *17*(4–5), 471–490. doi: 10.1016/S0167-9457(98)00010-4

Kaufer, D. S., Hayes, J. R., & Flower, L. (1986). Composing written sentences. *Research in the Teaching of English*, *20*(2), 121–140.

Kellogg, R.T. (1996). A model of Working Memory in writing. In C.M. Levy, & S. Ransdell (Eds.). *The science of writing: Theories, methods, individual differences and applications* (pp. 57-72). Mahwah, NJ: L. Erlbaum Associates.

Kessler, B. (2009). Statistical learning of conditional orthographic correspondences. *Writing Systems Research*, *1*(1), 19–34. doi: 10.1093/wsr/wsp004

Kessler, B., Pollo, T. C., Treiman, R., & Cardoso-Martins, C. (2013). Frequency analyses of prephonological spellings as predictors of success in conventional spelling. *Journal of Learning Disabilities*, *46*(3), 252–259. doi: 10.1177/0022219412449440

Kessler, B., & Treiman, R. (2003). Is English spelling chaotic? Misconceptions concerning its irregularity. *Reading Psychology*, *24*(3–4), 267–289. doi: 10.1080/02702710390227228

Kim, Y.-S. G., & Schatschneider, C. (2017). Expanding the developmental models of writing: A direct and indirect effects model of developmental writing (DIEW). *Journal of Educational Psychology*, *109*(1), 35–50. doi: 10.1037/edu0000129.Expanding

Kudo, M. F., Lussier, C. M., & Swanson, H. L. (2015). Reading disabilities in children: A selective meta-analysis of the cognitive literature. *Research in Developmental Disabilities*, *40*, 51–62. doi: 10.1016/j.ridd.2015.01.002

Landerl, K., & Moll, K. (2010). Comorbidity of learning disorders: Prevalence and familial transmission. *Journal of Child Psychology and Psychiatry*, *51*(3), 287–294. doi: 10.1111/j.1469-7610.2009.02164.x

Landerl, K., Ramus, F., Moll, K., Lyytinen, H., Lohvansuu, K., Donovan, M. O., … Schulte-Körne, G. (2013). Predictors of developmental dyslexia in European orthographies with varying complexity. *Journal of child psychology and psychiatry*, *54*(6), 686–694. doi: 10.1111/jcpp.12029

Langmaid, R. A., Papadopoulos, N., Johnson, B. P., Phillips, J. G., & Rinehart, N. J. (2014). Handwriting in children With ADHD. *Journal of Attention Disorders*, *18*(6), 504–510. doi: 10.1177/1087054711434154

Laszlo, J. I., & Bairstow, P. J. (1983). Kinaesthesis: Its measurement, training and
relationship to motor control. *The Quarterly Journal of Experimental Psychology*, *35*(2),
411–421. doi: 10.1080/14640748308402142

Laszlo, J. I., Bairstow, P. J., Bartrip, J., & Rolfe, Ursula, T. (1988). Clumsiness or perceptuo-
motor dysfunction? *Cognition and Action in Skilled Behaviour*, *55*, 293–310.

Learning Wales. (2014). *National literacy and numeracy framework*. Retrieved from
http://learning.gov.wales/resources/browse-all/nlnf/?lang=en

Lervåg, A., & Hulme, C. (2009). Rapid automatized naming (RAN) taps a mechanism that
places constraints on the development of early reading fluency. *Psychological science*,
20(8), 1040–8. doi: 10.1111/j.1467-9280.2009.02405.x

Lewis, M., Vance, A., Maruff, P., Wilson, P., & Cairney, S. (2008). Differences in motor
imagery between children with developmental coordination disorder with and without
the combined type of ADHD. *Developmental Medicine and Child Neurology*, *50*, 608–
612. doi: 10.1111/j.1469-8749.2008.03030.x

Levelt, W.J.M., 1989. *Speaking: From intention to articulation*. Cambridge, MA: MIT Press,

Li, K.-Y., Su, W.-J., Fu, H.-W., & Pickett, K. A. (2015). Kinesthetic deficit in children with
developmental coordination disorder. *Research in Developmental Disabilities*, *38*, 125–
133. doi: 10.1016/j.ridd.2014.12.013

Lingam, R., Hunt, L., Golding, J., Jongmans, M., & Emond, A. (2009). Prevalence of
developmental coordination disorder using the DSM-IV at 7 years of age: a UK
population–based study. *Pediatrics*, *123*(4), e693–e700. doi: 10.1542/peds.2008-1770

Lobier, M., & Valdois, S. (2015). Visual attention deficits in developmental dyslexia cannot
be ascribed solely to poor reading experience. *Nature Reviews Neuroscience*, *16*(4), 225.
doi: 10.1038/nrn3836

Longcamp, M., Velay, J., Berninger, V. W., & Richards, T. (2016). Neuroanatomy of
handwriting and related reading and writing skills in adults and children with and
without learning disabilities: French-American connections. *Pratiques. Linguistique,
Lttérature, Didactique*, 171–172. doi: 10.4000/pratiques.3175

Longcamp, M., Zerbato-Poudou, M. T., & Velay, J. L. (2005). The influence of writing
practice on letter recognition in preschool children: A comparison between handwriting
and typing. *Acta psychologica*, *119*(1), 67-79.

Lord, R., & Hulme, C. (1987). Perceptual judgements of normal and clumsy children.
Developmental Medicine and Child Neurology, *29*(2), 250–257.

Lord, R., & Hulme, C. (1988). Visual perception and drawing ability in clumsy and normal

children. *British Journal of Developmental Psychology*, *6*, 1–9.

MacCallum, R. C., Zhang, S., Preacher, K. J., Rucker, D. D. (2002). On the practice of dichotomization of quantitative variables. *Psychological Methods, 7*(1), 19-40. doi: 10.1037//1082-989X.7.1.19

Maldarelli, J. E., Kahrs, B. A., Hunt, S. C., & Lockman, J. J. (2015). Development of early handwriting: Visual-motor control during letter copying. *Developmental Psychology*, *51*(7), 1–10. doi: 10.1037/a0039424

Manly, T., Anderson, V., Nimmo-smith, I., Turner, A., Watson, P., & Robertson, I. H. (2001). The Differential Assessment of Children ' s Attention: The Test of Everyday Attention for Children (TEA-Ch), Normative Sample and ADHD Performance. *Journal of Child Psychology and Psychiatry*, *42*(8), 1065–1081. doi: 10.1017/S0021963001007909

Manly, T. Robertson, I. H., Anderson, V., Nimmo-Smith, I. (1998). Test of everyday attention for children (TEA-Ch) [Assessment instrument]. London, UK: Pearson.

Margolin, David, I., & Wing, A. M. (1983). Agraphia and micrographia: Clinical manifestations of motor programming and performance disorders. *Acta Psychologica*, *54*(1–3), 263–283. doi: 10.1016/0001-6918(83)90039-2

Martin, N. C., Piek, J., Baynam, G., Levy, F., & Hay, D. (2010). An examination of the relationship between movement problems and four common developmental disorders. *Human Movement Science*, *29*(5), 799–808. doi: 10.1016/j.humov.2009.09.005

Martin, N. C., Piek, J. P., & Hay, D. (2006). DCD and ADHD: A genetic study of their shared aetiology. *Human Movement Science*, *25*(1), 110–124. doi: 10.1016/j.humov.2005.10.006

Martinet, C., Valdois, S., & Fayol, M. (2004). Lexical orthographic knowledge develops from the beginning of literacy acquisition. *Cognition*, *91*(2), 11–22. doi: 10.1016/j.cognition.2003.09.002

Martlew, M. (1992). Handwriting and spelling: Dyslexic children's abilities compared with children of the same chronological age and younger children of the same spelling level. *British Journal of Educational Psychology*, *625*, 375–390.

Masterson, S.

McCutchen, D. (1996). A capacity theory of writing: Working memory in composition. *Educational Psychology Review*, *8*(3), 299–325. doi: 10.1007/BF01464076

McCutchen, D. (2000). Knowledge, processing, and working memory: Implications for a theory of writing. *Educational Psychologist*, *35*(1), 13–23.

McCutchen, D. (2011). From novice to expert: Implications of language skills and writing-relevant knowledge for memory during the development of writing skill. *Journal of Writing Research*, *3*(1), 51–68.

Mcgee, R., Brodeur, D., Symons, D., Andrade, B., & Fahie, C. (2004). Time perception: Does it distinguish ADHD and RD children in a clinical sample? *Journal of Abnormal Child Psychology*, *32*(5), 481–490. doi: 10.1023/B:JACP.0000037778.61929.1b

Mcintyre, F., Parker, H., Thornton, A., Licari, M., Piek, J., Rigoli, D., & Hands, B. (2017). Assessing motor proficiency in young adults: The Bruininks Oseretsky Test-2 Short Form and the McCarron Assessment of Neuromuscular Development. *Human Movement Science*, *53*, 55–62.

Melby-lervåg, M., & Lervåg, A. (2012). Oral language skills moderate nonword repetition skills in children with dyslexia: A meta-analysis of the role of nonword repetition skills in dyslexia oral language skills moderate nonword repetition skills in children with dyslexia: A meta-analysis of t. *Scientific Studies of Reading*, *16*(1), 1–34. doi: 10.1080/10888438.2010.537715

Melby-Lervåg, M., Lyster, S.-A., & Hulme, C. (2012). Phonological skills and their role in learning to read: A meta-analytic review. *Psychological Bulletin*, *138*(2), 322–352. doi: 10.1037/a0026744

Menghini, D., Finzi, A., Benassi, M., Bolzani, R., & Facoetti, A. (2010). Different underlying neurocognitive deficits in developmental dyslexia: A comparative study. *Neuropsychologia*, *48*(4), 863–865. doi: 10.1016/j.neuropsychologia.2009.11.003

Meulenbroek, R. G., & van Galen, G. P. (1986). Movement analysis of repetitive writing behavior of first, second and third grade primary school children. *Graphonomics: Contemporary Research in Handwriting*, *37*, 71–92. doi: 10.1016/S0166-4115(09)60073-X

Meulenbroek, R. G., & van Galen, G. P. (1990). Perceptual-motor complexity of printed and cursive letters. *Journal of Experimental Education*, *58*(2), 95–110. doi: 10.1080/00220973.1990.10806527

Milfont, T. L., & Fischer, R. (2010). Testing measurement invariance across groups: Applications in cross-cultural research. *International Journal of Psychological Research*, *3*(1), 111–121.

Miller, A. C., Fuchs, D., Fuchs, L. S., Compton, D. L., Kearns, D., Zhang, W., … Kircnner, D. (2014). Behavioral attention: A longitudinal study of whether and how it influences the development of word reading and reading comprehension among at-risk readers.

Journal of research on educational effectiveness, *7*(3), 232–249. doi: 10.1080/19345747.2014.906691.Behavioral

Miller, L. T., Missiuna, C. A., Macnab, J. J., Malloy-Miller, T., & Polatajko, H. J. (2001). Clinical description of children with developmental coordination disorder. *Canadian Journal of Occupational Therapy*, *68*(1), 5–15. doi: 10.1177/000841740106800101

Moll, K., Göbel, S. M., Gooch, D., Landerl, K., & Snowling, M. J. (2016). Cognitive Risk Factors for Specific Learning Disorder: Processing Speed, Temporal Processing, and Working Memory. *Journal of Learning Disabilities*, *49*(3), 272–281. doi: 10.1177/0022219414547221

Moll, K., & Landerl, K. (2009). Double dissociation between reading and spelling deficits. *Scientific Studies of Reading*, *13*(5), 359–382. doi: 10.1080/10888430903162878

Moll, K., Ramus, F., Bartling, J., Bruder, J., Kunze, S., Neuhoff, N., … Landerl, K. (2014). Cognitive mechanisms underlying reading and spelling development in five European orthographies. *Learning and Instruction*, *29*, 65–77. doi: 10.1016/j.learninstruc.2013.09.003

Mon-Williams, M. A., Wann, J., & Pascal, E. (1999). Visual–proprioceptive mapping in children with developmental coordination disorder. *Developmental Medicine and Child Neurology*, *41*, 247–254.

Muthén, L. K.., & Muthén, B. O. (2014). Mplus User's Guide (Seventh Edition). Los Angeles, CA: Muthén & Muthén.

Nicolson, R. I., & Fawcett, A. J. (1990). Automaticity: A new framework for dyslexia research? *Cognition*, *35*(2), 159–182.

Nicolson, R. I., & Fawcett, A. J. (1999). Developmental dyslexia: The role of the cerebellum. *Dyslexia*, *5*, 155–177.

Nicolson, R. I., & Fawcett, A. J. (2000). *Long-term learning in dyslexic children Long-term learning in dyslexic children* (Vol 12). doi: 10.1080/09541440050114552

Nicolson, R. I., & Fawcett, A. J. (2007). Procedural learning difficulties: Reuniting the developmental disorders? *Trends in Neurosciences*, *30*(4), 134–141. doi: 10.1016/j.tins.2007.02.003

Nicolson, R. I., & Fawcett, A. J. (2011). Dyslexia, dysgraphia, procedural learning and the cerebellum. *Cortex*, *47*(1), 117–127. doi: 10.1016/j.cortex.2009.08.016

Nicolson, R. I., Fawcett, A. J., & Dean, P. (2001). Developmental dyslexia: The cerebellar deficit hypothesis. *Trends in Neurosciences*, *24*(9), 508–511.

Olive, T. (2010). Methods, techniques, and tools for the on-line study of the writing process.

In N.L. Mertens (Ed.), Writing: processes, tools and techniques (pp. 1–18). New York, NY: Nova Science Publishers.

Olive, T. (2014). Toward a parallel and cascading model of the writing system: A Review of Research on Writing Processes Coordination. *Journal of Writing Research*, *6*(2), 173–194.

Olive, T., & Kellogg, R. T. (2002). Concurrent activation of high- and low-level production processes in written composition. *Memory & Cognition*, *30*(4), 594–600. doi: 10.3758/BF03194960

Palmis, S., Danna, J., Velay, J., & Longcamp, M. (2017). Motor control of handwriting in the developing brain: A review. *Cognitive Neuropsychology*, *34*(3–4), 187–204. doi: 10.1080/02643294.2017.1367654

Parush, S., Yochman, A., Cohen, D., & Gershon, E. (1998). Relation of visual perception and visual-motor integration for clumsy children. *Perceptual and Motor Skills*, *86*(1), 291–295.

Paz-Villagrán, V., Danna, J., & Velay, J.-L. (2014). Lifts and stops in proficient and dysgraphic handwriting. *Human Movement Science*, *33*, 381–394. doi: 10.1016/j.humov.2013.11.005

Pennington, B. F. (2006). From single to multiple deficit models of developmental disorders. *Cognition*, *101*(2), 385–413. doi: 10.1016/j.cognition.2006.04.008

Pennington, B. F., & Bishop, D. V. M. (2009). Relations among speech, language, and reading disorders. *Annual Review of Psychology*, *60*, 283–306. doi: 10.1146/annurev.psych.60.110707.163548

Pennington, B. F., Groisser, D., & Welsh, M. C. (1993). Contrasting cognitive deficits in attention deficit hyperactivity disorder versus reading disability. *Developmental Psychology*, *29*(3), 511–523.

Pennington, B. F., & Lefly, D. L. (2001). Early reading development in children at family risk for dyslexia. *Child Development*, *72*(3), 816–833.

Pick Jr., H. L., & Teulings, H.-L. (1983). Geometric transformations of handwriting as a function of instruction and feedback. *Acta Psychologica*, *54*(1–3), 327–340. doi: 10.1016/0001-6918(83)90044-6

Piek, J. P., Pitcher, T. M., & Hay, D. A. (1999). Motor coordination and kinaesthesis in boys with attention deficit – hyperactivity disorder. *Developmental Medicine and Child Neurology*, *41*(3), 159–165.

Polanczyk, G., De Lima, M. S., Horta, B. L., Biederman, J., & Rohde, L. A. (2007). The

worldwide prevalence of ADHD: A systematic review and metaregression analysis. *American Journal of Psychiatry*, *164*(6), 942–948.

Polatajko, Helene, J., Macnab, J. J., Anstett, B., Malloy-Miller, T., Murphy, K., & Noh, S. (1995). A clinical trial of the process-oriented treatment approach for children with developmental co-ordination disorder. *Developmental Medicine and Child Neurology*, *37*(4), 310–319.

Pontart, V., Bidet-Ildei, C., Lambert, E., Morisset, P., Flouret, L., & Alamargot, D. (2013). Influence of handwriting skills during spelling in primary and lower secondary grades. *Frontiers in Psychology*, *4*, 1–9. doi: 10.3389/fpsyg.2013.00818

Portier, Stanley, J., van Galen, G. P., & Meulenbroek, R. G. (1990). Practice and the dynamics of handwriting performance: Evidence for a shift of motor programming load. *Journal of Motor Behaviour*, *22*(4), 474–492. doi: 10.1080/00222895.1990.10735524

Prunty, M., & Barnett, A. L. (2017). Understanding handwriting difficulties: A comparison of children with and without motor impairment. *Cognitive Neuropsychology*, *34*(3–4), 205–218. doi: 10.1080/02643294.2017.1376630

Prunty, M., & Barnett, A. L. (in press). Accuracy and consistency of letter formation in children with Developmental Coordination Disorder: an exploratory study. *Journal of Learning Disabilities.*

Prunty, M., Barnett, A. L., Wilmut, K., & Plumb, M. (2016). Visual perceptual and handwriting skills in children with Developmental Coordination Disorder. *Human Movement Science*, *49*, 54–65. doi: 10.1016/j.humov.2016.06.003

Prunty, M., Barnett, A. L., Wilmut, K., & Plumb, M. S. (2013). Handwriting speed in children with Developmental Coordination Disorder: Are they really slower? *Research in developmental disabilities*, *34*(9), 2927–36. doi: 10.1016/j.ridd.2013.06.005

Prunty, M., Barnett, A. L., Wilmut, K., & Plumb, M. S. (2014). An examination of writing pauses in the handwriting of children with Developmental Coordination Disorder. *Research in Developmental Disabilities*, *35*(11), 2894–2905. doi: 10.1016/j.ridd.2014.07.033

Raberger, T., & Wimmer, H. (2003). On the automaticity/cerebellar deficit hypothesis of dyslexia: Balancing and continuous rapid naming in dyslexic and ADHD children. *Neuropsychologia*, *41*(11), 1493–1497. doi: 10.1016/S0028-3932(03)00078-2

Racine, M. B., Majnemer, A., Shevell, M., & Snider, L. (2008). Handwriting performance in children with attention deficit hyperactivity disorder (ADHD). *Journal of child neurology*, *23*(4), 399–406. doi: 10.1177/0883073807309244

Ramus, F., Marshall, C. R., Rosen, S., & van der Lely, H. K. J. (2013). Phonological deficits in specific language impairment and developmental dyslexia: Towards a multidimensional model. *Brain*, *136*(2), 630–645. doi: 10.1093/brain/aws356

Ramus, F., Pidgeon, E., & Frith, U. (2003). The relationship between motor control and phonology in dyslexic children. *Journal of Child Psychology and Psychiatry*, *44*(5), 712–722.

Ramus, F., & Szenkovits, G. (2008). What phonological deficit? *The Quarterly Journal of Experimental Psychology*, *61*(1), 129–141. doi: 10.1080/17470210701508822

Ransby, M. J., & Lee Swanson, H. (2003). Reading Comprehension Skills of Young Adults with Childhood Diagnoses of Dyslexia. *Journal of Learning Disabilities*, *36*(6), 538–555. doi: 10.1177/00222194030360060501

Rapp, B., Epstein, C., & Tainturier, M.-J. (2002). The integration of information across lexical and sublexical processes in spelling. *Cognitive Neuropsychology*, *19*(1), 1–29. doi: 10.1080/0264329014300060

Rasmussen, P., & Gillberg, C. (1996). Natural Outcome of ADHD With Developmental Coordination Disorder at Age 22 Years : A Controlled , Longitudinal , Community-Based Study. *Journal of the American Academy of Child & Adolescent Psychiatry*, *39*(11), 1424–1431. doi: 10.1097/00004583-200011000-00017

Rochelle, K. S. H., & Talcott, J. B. (2006). Impaired balance in developmental dyslexia? A meta-analysis of the contending evidence. *Journal of Child Psychology and Psychiatry*, *47*(11), 1159–66. doi: 10.1111/j.1469-7610.2006.01641.x

Romani, C., Olson, A., & Betta, A. D. M. (2005). Spelling Disorders The Dual-Route Model of Spelling. In M. J. Snowling & C. Hulme (Reds), *The Science of Reading: A Handbook* (1st ed, bll 618–641). Oxford: Wiley.

Rose, J. (2009). *Identifying and Teaching Children and Young People with Dyslexia and Literacy Difficulties Identifying and Teaching Children and Young People with Dyslexia and Literacy Difficulties*.

Rosenblum, S., Goldstand, S., & Parush, S. (2006). Relationships among biomechanical ergonomic factors, handwriting product quality, handwriting efficiency, and computerized handwriting process measures in children with and without handwriting difficulties. *American Journal of Occupational Therapy*, *60*(1), 28–39.

Rosenblum, S., & Livneh-Zirinski, M. (2008). Handwriting process and product characteristics of children diagnosed with developmental coordination disorder. *Human movement science*, *27*(2), 200–14. doi: 10.1016/j.humov.2008.02.011

Rosenblum, S., Margieh, J. A., & Engel-Yeger, B. (2013). Handwriting features of children with developmental coordination disorder - Results of triangular evaluation. *Research in Developmental Disabilities*, *34*(11), 4134–4141. doi: 10.1016/j.ridd.2013.08.009

Rosenblum, S., Weiss, P. L., & Parush, S. (2003). Product and process evaluation of handwriting difficulties: A review. *Educational Psychology Review*, *15*(1), 41–81.

Rothlein, D., & Rapp, B. (2014). The similarity structure of distributed neural responses reveals the multiple representations of letters. *NeuroImage*, *89*, 331–344. doi: 10.1016/j.neuroimage.2013.11.054.The

Roux, S., McKeeff, T. J., Grosjacques, G., Afonso, O., & Kandel, S. (2013). The interaction between central and peripheral processes in handwriting production. *Cognition*, *127*(2), 235–241. doi: 10.1016/j.cognition.2012.12.009

Rowe, D. W., & Wilson, S. J. (2015). The development of a descriptive measure of early childhood writing: Results from the Write Start! writing assessment. *Journal of Literacy Research*, *47*(2), 245–292. doi: 10.1177/1086296X15619723

Rucklidge, J. J., & Tannock, R. (2002). Neuropsychological profiles of adolescents with ADHD: Effects of reading difficulties and gender. *Journal of Child Psychology and Psychiatry*, *43*(8), 988–1003. doi: 10.1111/1469-7610.00227

Rutter, M., Fergusson, D., Horwood, L. J., Goodman, R., Maughan, B., Moffitt, T. E., … Page, P. (2004). Sex Differences in Developmental Reading Disability. *JAMA*, *291*(16), 2007–2012.

Sala, S. Della, & Cubelli, R. (2007). "Directional apraxia ": A unitary account of mirror writing following brain injury or as found in normal young children. *Journal of Neuropsychology*, *1*, 3–26. doi: 10.1348/174866407X180783

Santangelo, T., Harris, K., & Graham, S. (2016). Self-regulation and writing. *Handbook of writing research* (pp. 174-193).

Schoemaker, M. M., Lingam, R., Jongmans, M. J., van Heuvelen, M. J. G., & Emond, A. (2013). Is severity of motor coordination difficulties related to co-morbidity in children at risk for developmental coordination disorder ? *Research in Developmental Disabilities*, *34*(10), 3084–3091. doi: 10.1016/j.ridd.2013.06.028

Schoemaker, M. M., Wees, M. Van Der, Flapper, B., Verheij-Jansen, N., Scholten-Jaegers, S., & Geuze, R. H. (2001). Perceptual skills of children with developmental coordination disorder. *Human Movement Science*, *20*(1–2), 111–133.

Shapiro, M. B., Morris, R. D., Morris, M. K., & Jones, R. W. (1998). A neuropsychologically based assessment model of the structure of attention in children. *Developmental*

Neurospcyhology, *14*(4), 657–677. doi: 10.1080/87565649809540735

Shaywitz, S. E., Shaywitz, B. A., Fletcher, J. M., & Escobar, M. D. (1990). Prevelance of reading disability in boys and girls: Results of the Connecticut longitudinal study. *JAMA*, *264*(8), 998–1002.

Shepard, R. N., & Metzler, J. (1971). Mental rotation of three-dimensional objects. *Science*, *171*(3972), 701–704.

Sigmundsson, H., Hansen, P. C., & Talcott, J. B. (2003). Do "clumsy" children have visual deficits. *Behavioural Brain Research*, *139*, 123–129.

Simon, H. A., & Hayes, J. R. (1976). The understanding process: Problem isomorphs. *Cognitive Psychology*, *8*(2), 165–190. doi: 10.1016/0010-0285(76)90022-0

Smits-Engelsman, B., Schoemaker, M. M., Delabastita, T., Hoskens, J., & Geuze, R. (2015). Diagnostic criteria for DCD: Past and future. *Human Movement Science*, *42*, 293–306. doi: 10.1016/j.humov.2015.03.010

Smyth, M. M., & Mason, U. C. (1997). Planning and execution of action in children with and without developmental coordination disorder. *Journal of Child Psychology and Psychiatry*, *38*(8), 1023–1037.

Snowling, M. J. (2008). Specific disorders and broader phenotypes: The case of dyslexia. *The Quarterly Journal of Experimental Psychology*, *61*(1), 142–156. doi: 10.1080/17470210701508830

Snowling, M. J. (2013). Early identification and interventions for dyslexia: a contemporary view. *Journal of Research in Special Educational Needs*, *13*(1), 7–14. doi: 10.1111/j.1471-3802.2012.01262.x

Snowling, M. J., & Hulme, C. (2015). Disorders of reading, mathematical and motor development. In A. Thapar, D. S. Pine, J. F. Leckman, S. Scott, M. J. Snowling, & E. Taylor (Reds), *Rutter's Child and Adolescent Psychiarty* (6th ed, bll 702–719). Oxford: John Wiley & Sons.

Snowling, M. J., Stothard, S. E., Clarke, P., Bowyer-Crane, C., Harrington, A., Truelove, E., ... & Hulme, C. (2009). *YARC York Assessment of Reading for Comprehension Passage Reading*. GL Publishers..

Sprenger-Charolles, L., Siegel, L. S., Béchennec, D., & Serniclaes, W. (2003). Development of phonological and orthographic processing in reading aloud, in silent reading, and in spelling: A four-year longitudinal study. *Journal of Experimental Child Psychology*, *84*(3), 194–217. doi: 10.1016/S0022-0965(03)00024-9

Sprenger-Charolles, L., Siegel, L. S., & Bonnet, P. (1998). Reading and spelling acquisition

in French: The role of phonological mediation and orthographic factors. *Journal of Experimental Child Psychology*, *68*(2), 134–165. doi: 10.1006/jecp.1997.2422

Stainthorp, R., Barnett, A., Henderson, S., & Scheib, B. (2006). Handwriting, policy and practice in primary schools.

StataCorp. 2013. *Stata Statistical Software: Release 13*. College Station, TX: StataCorp LP.

Stein, J. F. (2001). The magnocellular theory of developmental dyslexia. *Dyslexia*, *7*(1), 12–36. doi: 10.1002/dys.186

Stein, J. F. (2018). Does dyslexia exist? *Language, Cognition and Neuroscience*, *33*(3), 313–320. doi: 10.1080/23273798.2017.1325509

Sumner, E., Connelly, V., & Barnett, A. L. (2013). Children with dyslexia are slow writers because they pause more often and not because they are slow at handwriting execution. *Reading and Writing*, *26*(6), 991–1008. doi: 10.1007/s11145-012-9403-6

Sumner, E., Connelly, V., & Barnett, A. L. (2014). The influence of spelling ability on handwriting production: Children with and without dyslexia. *Journal of Experimental Psychology. Learning, Memory, and Cognition*, *40*(5), 1441–7. doi: 10.1037/a0035785

Sumner, E., Connelly, V., & Barnett, A. L. (2016). The influence of spelling ability on vocabulary choices when writing for children with dyslexia. *Journal of Learning Disabilities*, *49*(3), 293–304. doi: 10.1177/0022219414552018

Sumner, E., Pratt, M. L., & Hill, E. L. (2016). Examining the cognitive profile of children with developmental coordination disorder. *Research in Developmental Disabilities*, *56*, 10–17. doi: 10.1016/j.ridd.2016.05.012

Swanson, H. L., & Berninger, V. W. (1996). Individual differences in children's working memory and writing skill. *Journal of Experimental Child Psychology*, *63*(2), 358–385.

Swanson, H. L., Zheng, X., & Jerman, O. (2009). Working memory, short-term memory, and reading disabilities: A selective meta-analysis of the literature. *Journal of Learning Disabilities*, *42*(3), 260–287. doi: 10.1177/0022219409331958

Teulings, H.-L., Thomassen, J. W. M., & van Galen, G. P. (1983). Preparation of partly precued handwriting movements: The size of the movment units in handwriting. *Acta Psychologica*, *54*(1–3), 165–177. doi: doi:10.1016/0001-6918(83)90031-8

Thapar, A., & Rutter, M. (2015). Neurodevelopmental disorders. In A. Thapar, D. S. Pine, J. F. Leckman, S. Scott, M. J. Snowling, & E. Taylor (eds), *Rutter's Child and Adolescent Psychiarty* (6th ed, bll 31–40). Oxford: John Wiley & Sons.

Thibon, L. S., Gerber, S., & Kandel, S. (2018). The elaboration of motor programs for the automation of letter production. *Acta Psychologica*, *182*, 200–211. doi:

10.1016/j.actpsy.2017.12.001

Thompson, P. a., Hulme, C., Nash, H. M., Gooch, D., Hayiou-Thomas, E., & Snowling, M. J. (2015). Developmental dyslexia: Predicting individual risk. *Journal of Child Psychology and Psychiatry*, *56*(9), 976–987. doi: 10.1111/jcpp.12412

Tolchinsky, L., Liberman, G., & Alonso-Cortes Fradejas, M. D. (2015). Explaining first graders' achievements in spellings and word seperation in shallow orthographies. *Journal of Writing Research*, *6*(3), 279–361.

Torrance, M., & Galbraith, D. (2006). The processing demands of writing. *Handbook of writing research* (pp. 67-80).

Treiman, R. (2017a). Learning to spell: Phonology and beyond. *Cognitive Neuropsychology*, *34*(3–4), 83–93. doi: 10.1080/02643294.2017.1337630

Treiman, R. (2017b). Learning to spell words: Findings, theories, and issues learning to spell words: Findings, theories, and issues. *Scientific Studies of Reading*, *34*(3–4), 83–93. doi: 10.1080/10888438.2017.1296449

Treiman, R., Decker, K., Kessler, B., & Pollo, T. C. (2015). Variation and repetition in the spelling of young children. *Journal of Experimental Child Psychology*, *132*, 99–110. doi: 10.1016/j.jecp.2014.12.008

Treiman, R., Gordon, J., Boada, R., Peterson, R. L., Pennington, B. F., Gordon, J., … Gordon, J. (2014). Statistical learning, letter reversals, and reading. *Scientific Studies of Reading*, *18*(6), 383–394. doi: 10.1080/10888438.2013.873937

Treiman, R., & Kessler, B. (2011). Similarities among the shapes of writing and their effects on learning. *Written Language and Literacy*, *14*(1), 39–57. doi: 10.1075/wll.14.1.03tre

Treiman, R., & Kessler, B. (2014). *How children learn to write words*. UK: Oxford University Press.

Treiman, R., Kessler, B., & Caravolas, M. (2018). What methods of scoring young children's spelling best predict later spelling performance? *Journal of Research in Reading*. doi: 10.1111/1467-9817.12241

Treiman, R., Kessler, B., Pollo, T. C., Byrne, B., & Olson, R. K. (2016). Measures of kindergarten spelling and their relations to later spelling performance. *Scientific Studies of Reading*, *20*(5), 349–362. doi: 10.1080/10888438.2016.1186168

Treiman, R., Levin, I., & Kessler, B. (2012). Linking the shapes of alphabet letters to their sounds: The case of Hebrew. *Reading and Writing*, *25*(2), 569–585. doi: 10.1007/s11145-010-9286-3

Treiman, R., & Yin, L. (2011). Early differentiation between drawing and writing in Chinese

children. *Journal of Experimental Child Psychology*, *108*(4), 786–801. doi: 10.1016/j.jecp.2010.08.013

Tsai, C.-L., Wilson, P. H., & Wu, S. K. (2008). Role of visual–perceptual skills (non-motor) in children with developmental coordination disorder. *Human Movement Science*, *27*(4), 649–664. doi: 10.1016/j.humov.2007.10.002

Tsai, C.-L., & Wu, S.-K. (2008). Relationship of visual perceptual deficit and motor impairment in children with developmental coordination disorder. *Perceptual and Motor Skills*, *107*(2), 457–472. doi: 10.2466/PMS107.2.457-472

Tseng, M. H., & Chow, S. M. K. (2000). Perceptual-motor function of school-age children with slow handwriting speed. *American Journal of Occupational Therapy*, *54*, 83–88.

Tseng, M. H., & Murray, Elizabeth, A. (1994). Differences in perceptual-motor measures in children With good and poor handwriting. *The Occupational Therapy Journal of Research*, *14*(1), 19–36.

Vaessen, A., Gerretsen, P., & Blomert, L. (2009). Naming problems do not reflect a second independent core deficit in dyslexia: Double deficits explored. *Journal of Experimental Child Psychology*, *103*(2), 202–221. doi: 10.1016/j.jecp.2008.12.004

Valyear, K. F., Culham, J. C., Sharif, N., Westwood, D., & Goodale, M. A. (2006). A double dissociation between sensitivity to changes in object identity and object orientation in the ventral and dorsal visual streams: A human fMRI study. *Neuropsychologia*, *44*(2), 218–228. doi: 10.1016/j.neuropsychologia.2005.05.004

van Bergen, E., van der Leij, A., & de Jong, P. F. (2014). The intergenerational multiple deficit model and the case of dyslexia. *Frontiers in Human Neuroscience*, *8*(346), 1–13. doi: 10.3389/fnhum.2014.00346

van den Boer, M., & de Jong, P. F. (2018). Stability of visual attention span performance and its relation with reading over time. *Scientific Studies of Reading*, *22*(5), 434–441. doi: 10.1080/10888438.2018.1472266

van den Boer, M., van Bergen, E., & de Jong, P. F. (2015). The specific relation of visual attention span with reading and spelling in Dutch. *Learning and Individual Differences*, *39*, 141–149. doi: 10.1016/j.lindif.2015.03.017

van der Sluis, S., van der Leij, A., & de Jong, P. F. (2005). Working memory in Dutch children with reading- and arithmetic-related LD. *Journal of Learning Disabilities*, *38*(3), 207–221.

van Doorn, R. R. A., & Keuss, P. J. G. (1992). The role of vision in the temporal and spatial control of handwriting. *Acta Psychologica*, *81*, 269–286.

van Galen, G. P. (1991). Handwriting: psychomotor Issues for a theory. *Human Movement Science*, *10*, 165–191.

van Galen, G. P., & Teulings, H.-L. (1983). The independent monitoring of form and scale factors in handwriting. *Acta Psychologica*, *54*(1–3), 9–22. doi: 10.1016/0001-6918(83)90020-3

van Mier, H., & Hulstijn, W. (1993). The effects of motor complexity and practice on initiation time in writing and drawing. *Acta Psychologica*, *84*, 231–251.

Van Waelvelde, H., De Weerdt, W., De Cock, P., & Smits-engelsman, B. C. M. (2004). Association between visual perceptual deficits and motor deficits in children with developmental coordination disorder. *Developmental Medicine and Child Neurology*, *46*(10), 661–666.

Vandenberg, R. J., & Lance, C. E. (2000). A review and synthesis of the measurement invariance literature: Suggestions practices and recommendations for organizational research. *Organizational Research Methods*, *3*(1), 4–70.

Vander Hart, N., Fitzpatrick, P., & Cortesa, C. (2010). In-depth analysis of handwriting curriculum and instruction in four kindergarten classrooms. *Reading and Writing*, *23*(6), 673–699. doi: 10.1007/s11145-009-9178-6

Varvara, P., Varuzza, C., Sorrentino, Anna, C. P., Vicari, S., & Menghini, D. (2014). Executive functions in developmental dyslexia. *Frontiers in Human Neuroscience*, *8*(120), 1–8. doi: 10.3389/fnhum.2014.00120

Vellutino, F. R., Fletcher, J. M., Snowling, M. J., & Scanlon, D. M. (2004). Specific reading disability (dyslexia): What have we learned in the past four decades ? *Journal of Child Psychology and Psychiatry*, *45*(1), 2–40.

Vellutino, F. R., Scanlon, D. M., & Spearing, D. (1995). Semantic and phonological coding in poor and normal readers. *Journal of Experimental Child Psychologyerimental child psychology*, *59*(1), 76–123.

Visser, J., & Geuze, R. H. (2000). Kinaesthetic acuity in adolescent boys: A longitudinal study. *Developmental Medicine and Child Neurology*, *42*(2), 93–96.

Volman, M. J., van Schendel, B. M., & Jongmans, M. J. (2006). Handwriting difficulties in primary school childre: A search for underlying mechanisms. *The American Journal of Occupational Therapy*, *60*(4), 451–460. doi: 10.5014/ajot.60.4.451

Weintraub, N., & Graham, S. (2000). The contribution of gender, orthographic, finger function, and visual-motor processes to the prediction of handwriting status. *The Occupational Therapy Journal of Research*, *20*(2), 121–140.

Wechsler, D., Kaplan, E., Fein, D., Kramer, J., Morris, R., Delis, D., & Maelender, A. (2003). Wechsler intelligence scale for children: Fourth edition (WISC-IV) [Assessment instrument]. London, UK; Pearson.

Welsh Government. (2007) *Defining schools according to Welsh medium provision*. Retrieved from https://gov.wales/topics/educationandskills/publications/guidance/defining-schools-welsh-medium/?lang=en

Wilkinson, G. S., & Robertson, G. J. (2006). The Wide Range Achievement Test–4 (WRAT-4) [Assessment instrument]. Lutz, FL: Psychological Assessment Resources.

Willcutt, E. G., & Pennington, B. F. (2000). Comorbidity of reading disability and attention-deficit/hyperactivity disorder: Differences by gender and subtype. *Journal of Learning Disabilities*, *33*(2), 179–191.

Willcutt, E. G., Pennington, B. F., Boada, R., Ogline, J. S., Tunick, R. A., Chhabildas, N. A., & Olson, R. K. (2001). A comparison of the cognitive deficits in reading disability and attention-deficit/hyperactivity disorder. *Journal of Abnormal Psychology*, *110*(1), 157–172. doi: 10.1037//0021-843X.110.1.157

Willcutt, E. G., Pennington, B. F., Olson, R. K., Chhabildas, N., & Hulslander, J. (2005). Neuropsychological Analyses of Comorbidity Between Reading Disability and Attention Deficit Hyperactivity Disorder : In Search of the Common Deficit. *Developmental Neuropsychology*, *27*(1), 35–78.

Willcutt, E. G., Pennington, B. F., Smith, S. D., Cardon, L. R., Gaya, J., Knopik, V. S., … Defries, J. C. (2002). Quantitative trait locus for reading disability on chromosome 6p is pleiotropic for attention-deficit/hyperactivity disorder. *America Journal of Medical Genetics*, *114*(3), 260–268. doi: 10.1002/ajmg.10205

Williams, J., Anderson, V., Reddihough, D. S., Reid, S. M., Vijayakumar, N., & Wilson, P. H. (2011). A comparison of motor imagery performance in children with spastic hemiplegia and developmental coordination disorder. *Journal of Clinical and Experimental Neuropsychology*, *33*(3), 273–282. doi: 10.1080/13803395.2010.509714

Williams, J., Thomas, P. R., Maruff, P., & Wilson, P. (2008). The link between motor impairment level and motor imagery ability in children with developmental coordination disorder. *Human Movement Science*, *27*(2), 270–285. doi: 10.1016/j.humov.2008.02.008

Wilson, B. N., Kaplan, B. J., Crawford, S. G., Campbell, A., & Dewey, D. M. (2000). Reliability and validity of a parent questionnaire on childhood motor skills. *American Journal of Occupational Therapy*, *54*(5), 484–493. doi: 10.5014/ajot.54.5.484

Wilson, P. H., & Maruff, P. (1999). Deficits in the endogenous control of covert visuospatial attention in children with developmental coordination disorder. *Human Movement Science*, *18*(2–3), 421–442.

Wilson, P. H., Maruff, P., Butson, M., Williams, J., Lum, J., & Thomas, P. R. (2004). Internal representation of movement in children with developmental coordination disorder: A mental rotation task. *Developmental Medicine and Child Neurology*, *46*, 754–759.

Wilson, P. H., & McKenzie, B. E. (1998). Information Processing Deficits Associated with Developmental Coordination Disorder : A Meta-analysis of Research Findings. *Journal of Child Psychology and Psychiatry*, *39*, 829–840.

Wilson, P. H., Smits-Engelsman, B. C. M., Caeyenberghs, K., Steenbergen, B., Sugden, D. A., Clark, J., … Blank, R. (2017). Cognitive and neuroimaging findings in developmental coordination disorder: New insights from a systematic review of recent research. *Developmental Medicine and Child Neurology*, *59*(11), 1117–1129. doi: 10.1111/dmcn.13530

Wilson, P. H., Thomas, P. R., & Maruff, P. (2002). Motor imagery training ameliorates motor clumsiness in Children. *Journal of Child Neurology*, *17*(7), 491–498.

Wilson, P., Maruff, P., & McKenzie, B. E. (1997). Wilson, P. H., Maruff, P., & McKenzie, B. E. (1997). Covert orienting of visuospatial attention in children with developmental coordination disorder. *Developmental Medicine and Child Neurology*, *39*(11), 736–745.

Wilson, P., Ruddock, S., Smits-Engelsman, B., Polatajko, H., & Blank, R. (2013). Understanding performance deficits in developmental coordination disorder: A meta-analysis of recent research. *Developmental Medicine and Child Neurology*, *55*(3), 217–228. doi: 10.1111/j.1469-8749.2012.04436.x

Wimmer, H., Mayringer, H., & Landerl, K. (1998). Poor reading: A deficit in skill-automatization or a phonological deficit? *Scientific Studies of Reading*, *2*(4), 321–340. doi: 10.1207/s1532799xssr0204

Wimmer, H., Mayringer, H., & Raberger, T. (1999). Reading and dual-task balancing: Evidence against the automatization deficit explanation of developmental dyslexia. *Journal of Learning Disabilities*, *32*(5), 473–478.

Wolff, U. (2016). Effects of a randomized reading intervention study aimed at 9-Year-olds : A 5-year follow-up. *Dyslexia*, *22*(2), 85–100. doi: 10.1002/dys.1529

Wolpert, D. M. (1997). Computational approaches to motor control. *Trends in Cognitive Sciences*, *1*(6), 209–216.

Wolpert, D. M., & Ghahramani, Z. (2000). Computational principles of movement neuroscience. *Nature Neuroscience*, *3*(11s), 1212.

Wright, H. C., & Sugden, D. A. (1996). The nature of developmental coordination disorder: Inter- and intragroup differences. *Adapted Physical Activity Quarterly*, *13*(4), 357–371.

Yu, S.-H., & Chang, N.-Y. (2010). Characterization of motor control in handwriting difficulties in children with or without developmental coordination disorder. *Developmental Medicine and Child Neurology*, *52*(3), 244–250. doi: 10.1111/j.1469-8749.2009.03478.x

Zwicker, J. G., Missiuna, C., Harris, S. R., & Boyd, L. A. (2012). Developmental coordination disorder: A review and update. *European Journal of Paediatric Neurology*, *16*(6), 573–581. doi: 10.1016/j.ejpn.2012.05.005

Appendix A: Administration of Measures at Phases One, Two, and Three

Table A.1.

Order of Administration of Tests at Phases One, Two, and Three

Phase	Study	Test
1	1	WRIT Matrices
		Welsh Picture Word Matching
		Welsh Spelling
		WISC Coding
		Cloze Reading
	1+2[a]	WRAT Spelling
		Sentence Spelling
		Beery Visual Motor Integration
	1+3[a]	SaHLT Handwriting
2	2	Beery Visual Perception
		Phoneme Deletion
		RAN Objects
		WISC Digit Span
		One Minute Non-Word Reading
		WRAT Reading
		RAN Digits
		Beery Motor Coordination
		WMTB-C Corsa Block Task
		RAN Letters
		TEA-Ch Sky Search
		TEA-Ch Score!
		TEA-Ch Sky Search DT
		M-ABC 2 Bag Throw
		M-ABC 2 Board Balance
	3	Alphabet Writing
3	2	WISC Block Design
		M-ABC 2 Trail Drawing
		M-ABC 2 Threading
		One Minute Word Reading
	4	Novel Character Learning

Note. Order of tests presented per phase not match the exact order of administration.
[a]Data collected at Phase 1 was used in Study 1 and Study 2 or Study 3.

Appendix B: Single Group Confirmatory Factor Analysis Models
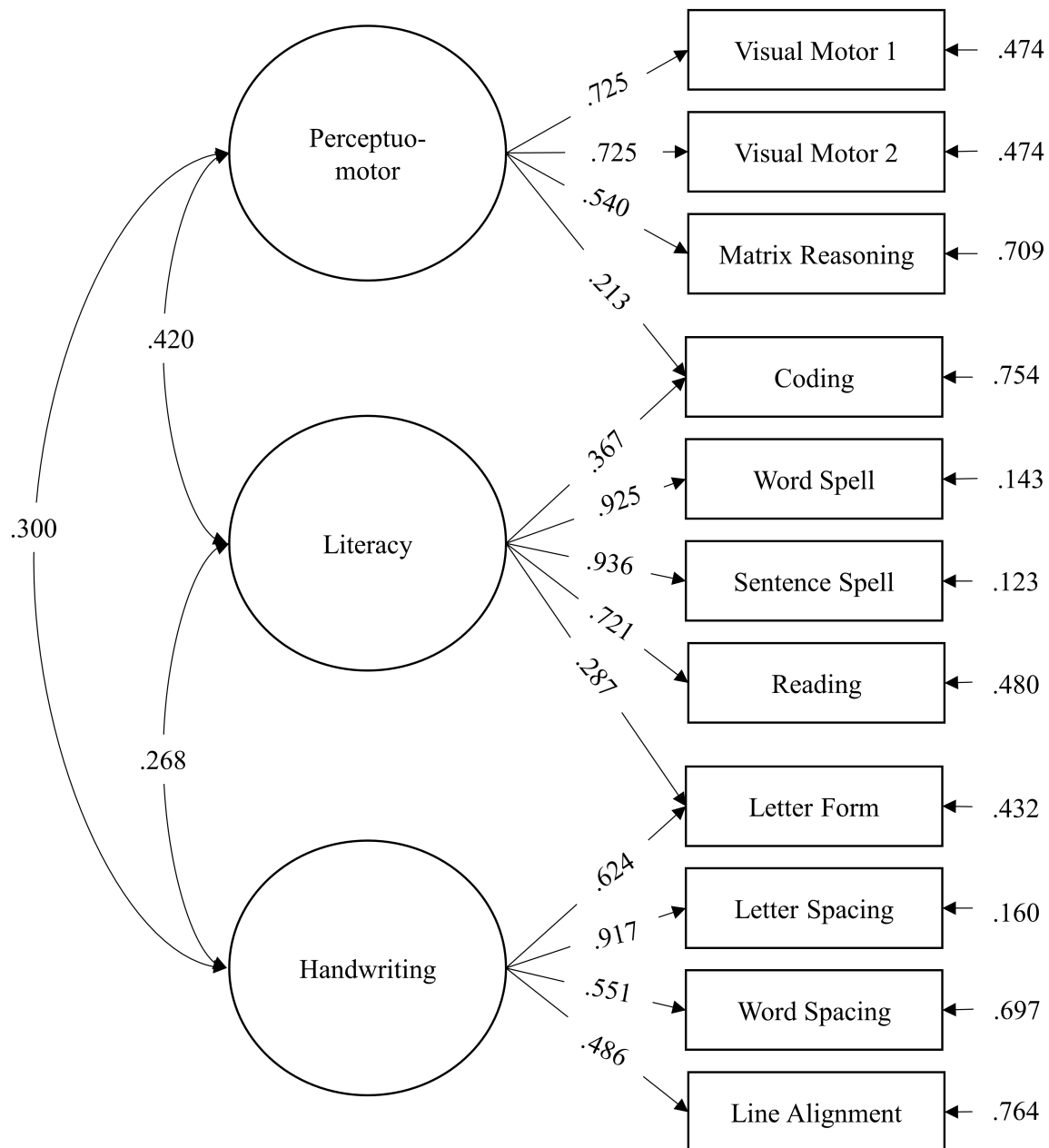
Figure B.1. Confirmatory factor analysis examined the factor structure of the class screening battery in Year 3. In total, 11 measures of literacy and motor skills were loaded onto three factors. Standardised path estimates with their residual variances are reported.
Visual Motor = Visual Motor Integration
All factor loadings were significant at *p* < .01.
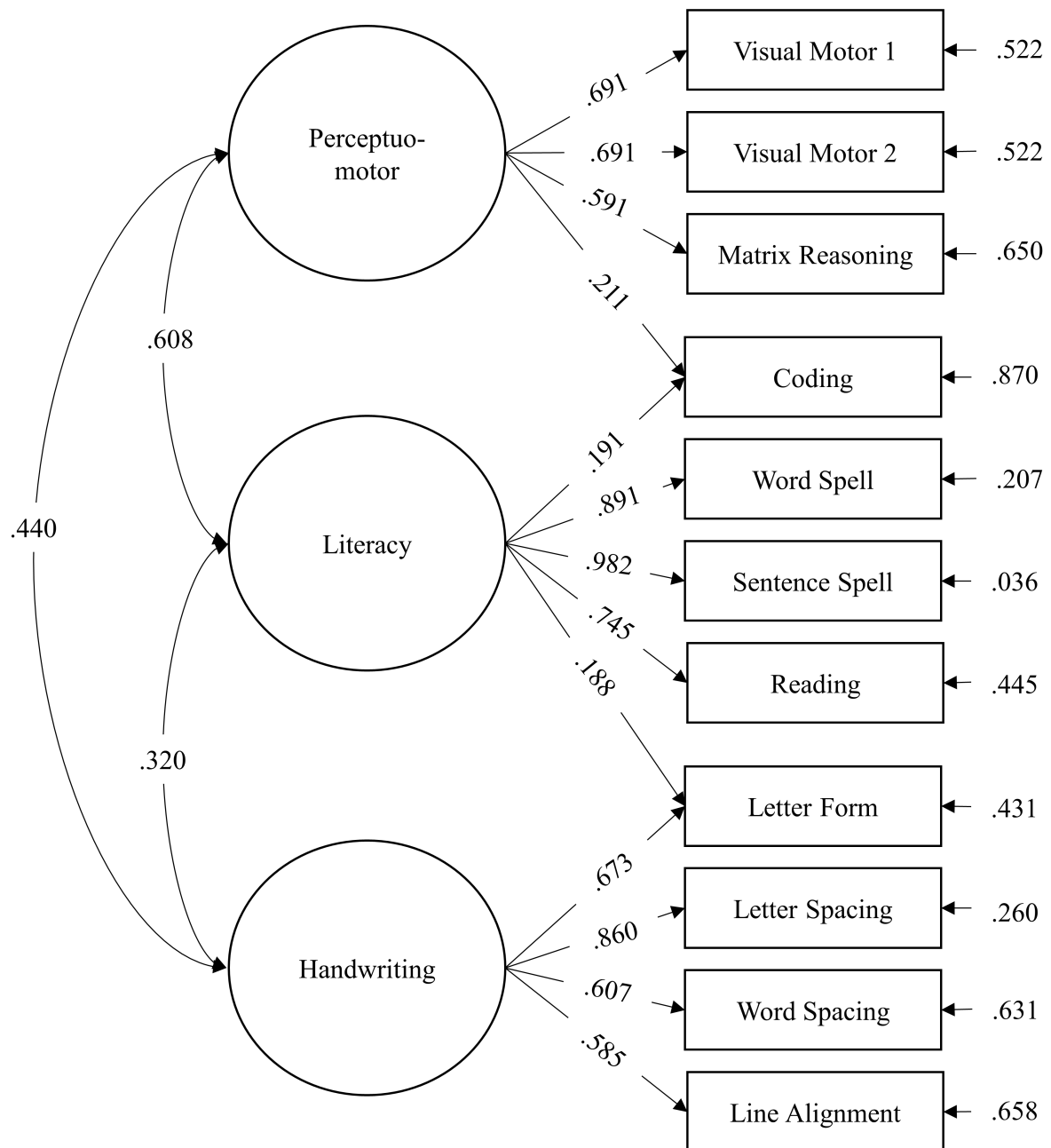
Figure B.2. Confirmatory factor analysis examined the factor structure of the class screening battery in Year 4. In total, 11 measures of literacy and motor skills were loaded onto three factors. Standardised path estimates with their residual variances are reported.

Visual Motor = Visual Motor Integration

All factor loadings were significant at $p < .05$.

Figure B.3. Confirmatory factor analysis examined the factor structure of the class screening battery in Year 5. In total, 11 measures of literacy and motor skills were loaded onto three factors. Standardised path estimates with their residual variances are reported.

Visual Motor = Visual Motor Integration
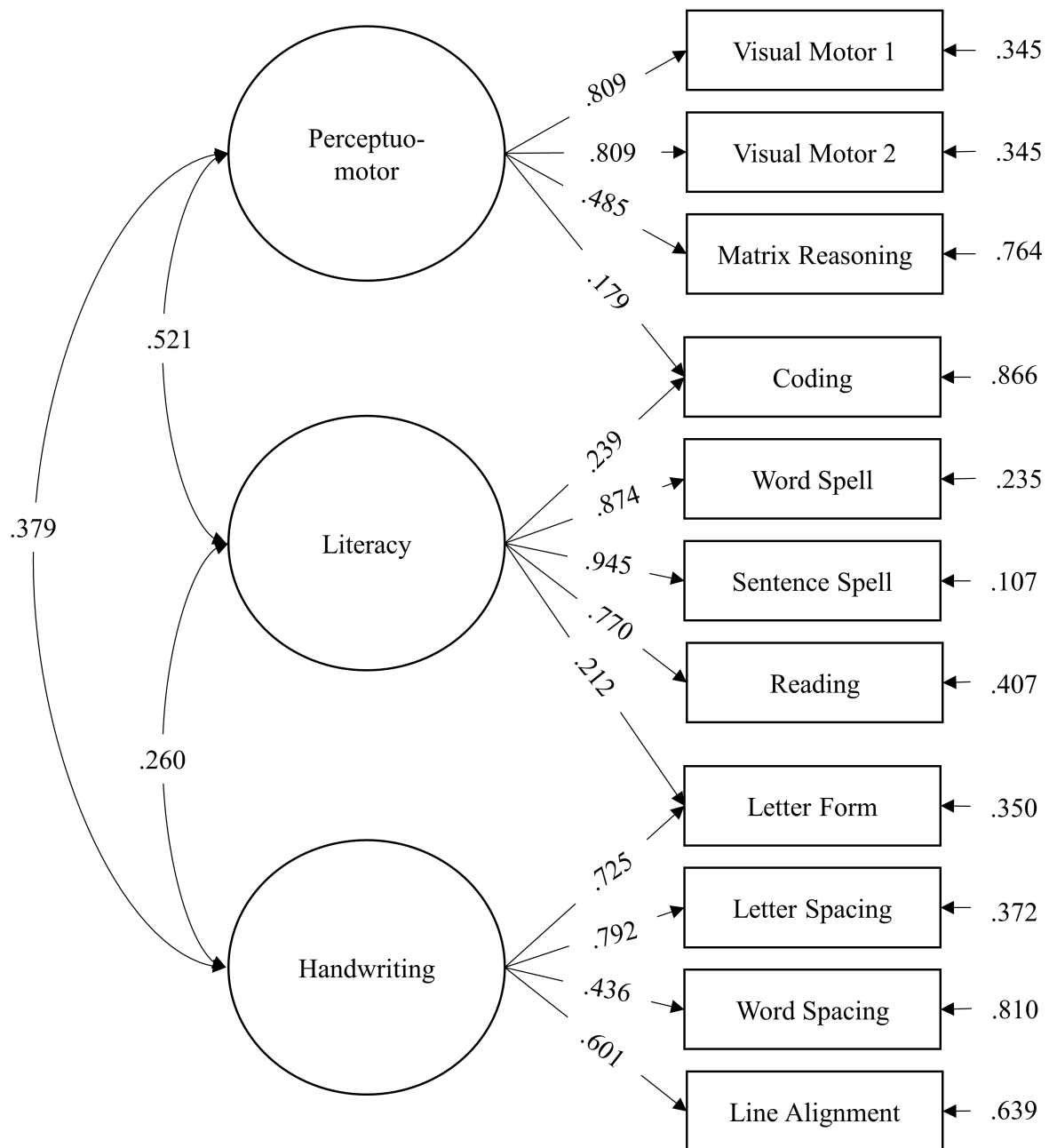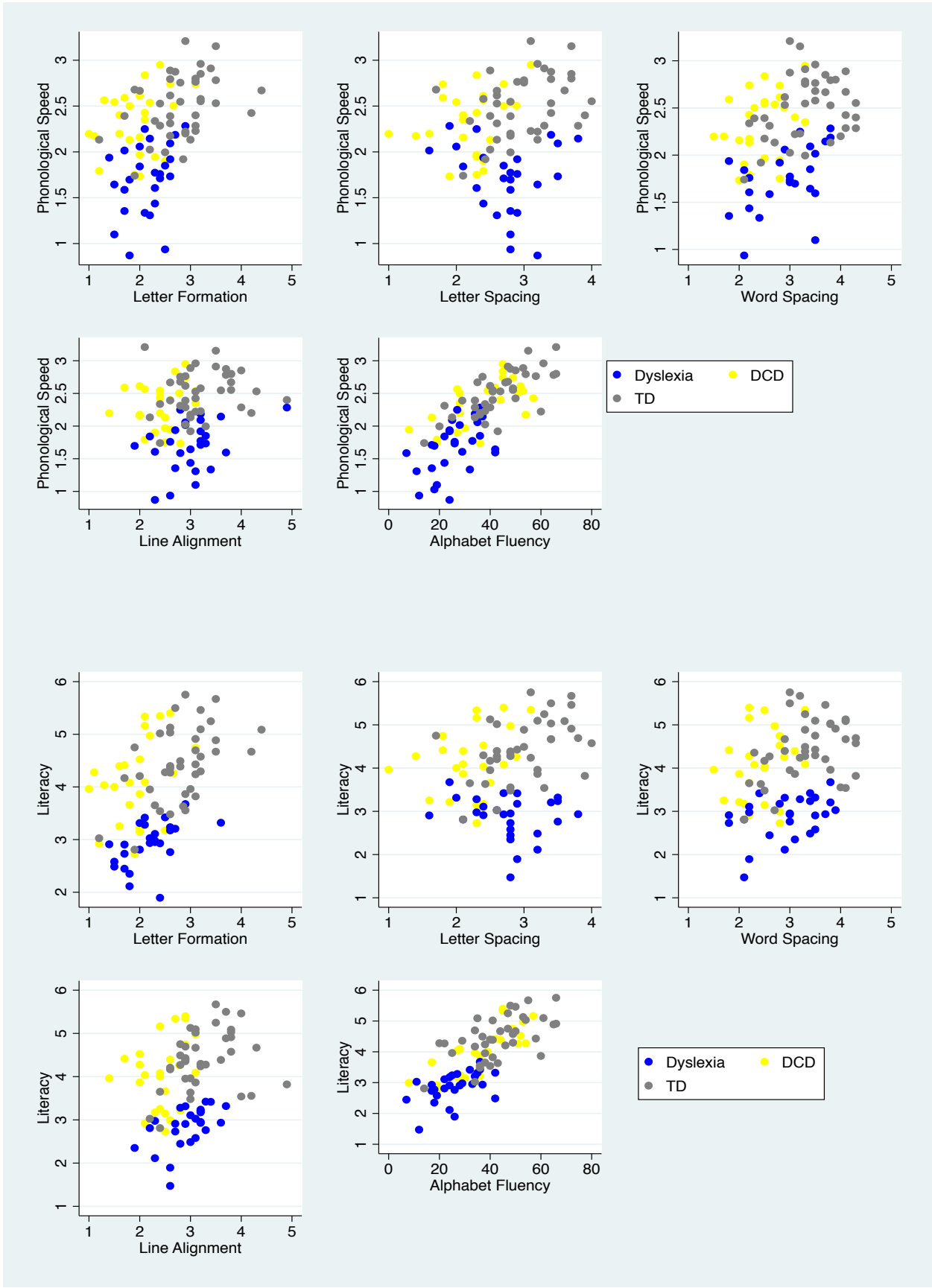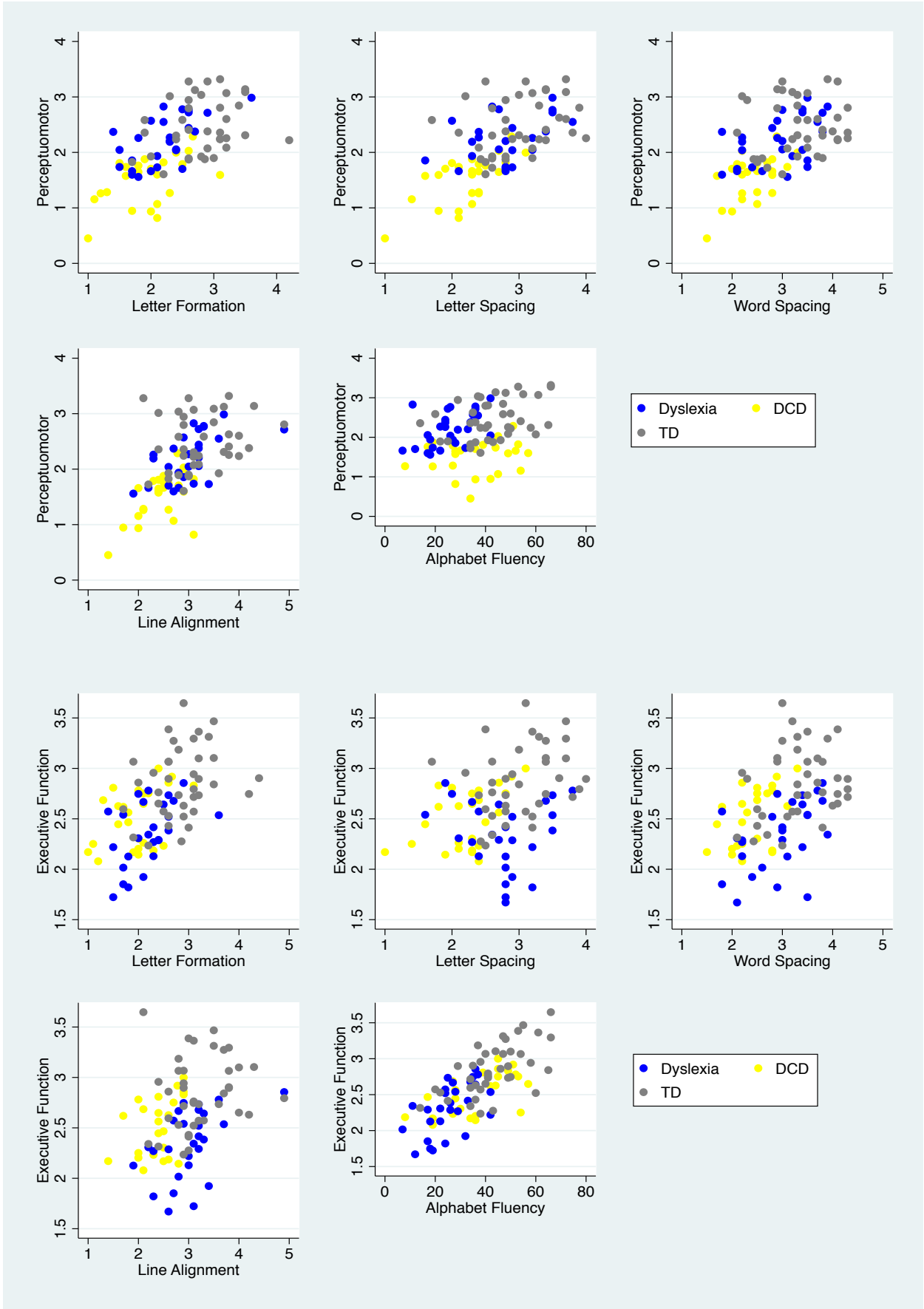
All factor loadings were significant at $p \leq .05$.

Appendix C: Scatter Plots of Handwriting Related Skills and Handwriting Legibility and Fluency

Appendix D: A Monte Carlo Power Analysis of a Simple Path Model

A Monte Carlo study was used to assess whether the current sample size ($N = 92$) was adequate for a simple two-path model using literacy and perceptuomotor factor score estimates to directly predict handwriting fluency. This analysis formed the first part of a stepwise approach, whereby this simple model was a baseline model to test if there was adequate power in a straight forward model. If power was adequate, further, more complex models would be tested and eventually used to examine predictors of handwriting fluency and legibility in dyslexia and DCD.

The simulation was run with 92 observations and hypothesised parameters were based on previous studies of contributions of literacy and perceptuomotor factor score estimates to handwriting fluency (e.g., Abbott & Berninger, 1993; Study 2). Handwriting fluency was used rather than handwriting legibility as there is a larger literature from which to draw parameter estimates from (see Study 3). It was assumed that was no missing data and all variables were continuous and normally distributed.

The adequacy of the sample size was evaluated using the criteria proposed by Muthén & Muthén (2002). These criteria state (a) the parameter and standard error biases must not be larger than 10% of any parameter in the model, (b) standard error bias of the parameter being assessed are no greater than 5% of the parameter, (c) coverage estimates should be between .91 - .98, (d) power should be greater than .8. Applying these criteria to results of the Monte Carlo simulation presented in Table C.1 revealed that, in the main, parameter, standard error biases, and coverage met the criteria in the main. However, the power estimates for the perceptuomotor path (.397) was substantially below the recommended power of .8. This means that the current sample was not large enough to detect significant effects on some of the paths in this simple model and so this model was not used, and so more complex models were not tested.

Table C.1.

Parameter Bias, Standard Error Bias, and Power Estimates for a Two-Path Model of Literacy and Perceptuomotor Skills on Handwriting Fluency

	Estimates							
	Starting	Average	Parameter Bias	*SD*	*SE* Average	Standard Error Bias	Coverage	Power
Handwriting on								
Literacy	.300	.3021	.0070	.0804	.0783	-.0261	.944	.968
Perceptuomotor	.200	.1969	-.0155	.1263	.1173	-.0713	.938	.397
Literacy with Perceptuomotor	.320	.3176	-.0075	.0664	.0645	-.0286	.939	1.000
Means								
Literacy	3.860	3.8637	.0010	.0956	.0926	-.0314	.945	1.000
Perceptuomotor	2.124	2.1222	-.0008	.0625	.0618	-.0112	.948	1.000
Intercepts								
Handwriting	36.087	36.0839	-.0001	.2594	.2595	.0004	.944	1.000
Residual Variances								
Handwriting	.300	.2901	-.0330	.0437	.0425	-.0275	.908	1.000
Variances								
Literacy	.807	.8021	-.00061	.1229	.1176	-.0431	.931	1.000
Perceptuomotor	.361	.3573	-.0102	.0512	.0524	.0234	.940	1.000

Appendix E: Character Rating Results

Participants

A total of 30 students (*M* age = 26.5 years, 80% female) completed the questionnaire. Of the 30 participants, one person did not complete the motor complexity section and another participant's visual orientation ratings were removed as many their responses were 'neutral', including for those letters with a clear orientation (e.g., < b >) suggesting they did not understand/attend to the task. This left 29 participant's ratings for both visual orientation and motor complexity.

Results

Tables 1 and 2 report each the ratings for alphabet letters and novel characters, respectively. The column headed *category* is the orientation and motor complexity rating for each item given by the candidate. The column headed *participant rating* is the orientation and motor complexity rating with the highest percentage agreement from the participant raters for each item. The letter (b, d, n) in the column refers to the orientation of the item whereas the number (1, 2, 3, 4) refers to the number of strokes in the character. The breakdown of percentage agreement for each possible visual orientation and motor complexity option under the header *% participant agreement*. Where disagreement exists, the specific area/s of disagreement (visual orientation/motor complexity) are highlighted in red.

Alphabet Characters. Table E.1 shows alphabet letters were judged by the participants to have the same orientation as the candidates, in the main. However, there was some disagreement between the candidate and participant raters on the motor complexity of letters. The participants judged most letters to have two-strokes, and only identified three letters as having a three-stroke structure. The letters where most disagreement occurs all share a curve at the top of bottom of the stem. It is likely that the raters, as experienced writers have treated this curve as a continuation of the stroke which composes the stem,

therefore treating the stem and the curve as one stroke. Considering this disagreement between the original categorisation and participant's ratings it is important to re-visit the rationale for identifying curves as separable strokes, or segments, of letters.

Including the curve as a separate stroke is important in distinguishing more complex letters from less complex letters. For example, < j > is distinguished from < i > by the additional curve at the end of the stem. Additional evidence for including the curve as an additional stroke comes from the motor control literature. Although, the literature examining the effect of motor complexity of letters is sparse with no agreement on what defines the complexity of letters. Meulenbroek and Van Galen (1986, 1988, 1990) examined the effects of different types of cursive letter strokes on children's writing velocity. They found letters with straight lines were written more quickly than those with increased curvature because letters with ascenders or descenders can be written with higher velocities than curves (Meulenbroek & van Galen, 1990). Therefore, letters that include curved segments are likely to take longer to complete than letters without a curved segment, for example, the letter < i > is differentiated from < j > by the addition of a curved segment, and the inclusion of the curve increases the complexity of the letter. Given that the letters containing curves have lower velocity than letters containing a straight lines and curves distinguish between letters (e.g., < i > and < j >), they should be identified as a separate stroke.

Table E.1.

Ratings of the Visual Orientation and Motor Complexity of Alphabet Letters by the Candidate and Participant Raters

Item	Category	Participant Rating	% Participant Agreement						
			Orientation			Complexity			
			d	n	b	1	2	3	4
a	2d	2d	86	3	10	0	93	3	3
b	2b	2b	3	0	97	0	93	3	3
c	1b	1b	7	3	86	90	10	0	0
d	2d	2d	90	0	10	0	93	3	3
e	2b	2b	0	3	97	0	97	3	0
f	3b	2b	0	0	100	0	86	14	0
g	3d	2d	86	0	14	0	69	28	3
h	2b	2b	0	7	93	0	90	10	0
i	2n	2n	0	93	3	3	97	0	0
j	3d	2d	97	0	3	3	76	21	0
k	3b	3b	0	3	97	0	0	100	0
m	4b	3n	3	52	45	0	3	86	10
n	2b	2b	3	28	69	0	90	10	0
o	1n	1n	0	100	0	100	0	0	0
p	2b	2b	7	0	93	0	90	3	3
q	3d	2d	90	0	10	0	93	3	3
r	2b	2b	0	0	100	0	100	0	0
s	2d	1b	0	48	52	79	14	7	0
t	3b	2b	0	0	100	0	76	24	0
u	2d	2d	55	38	7	0	86	14	0
v	2b	2n	0	93	3	3	97	0	0
w	4n	4n	0	97	3	0	3	0	97
x	2n	2n	0	97	3	0	100	0	0
y	3d	2d	79	7	14	0	76	17	7
z	3d	3d	52	45	3	3	0	97	0

Novel Characters. The same colour identifiers were applied to the novel symbols in Table E.2. For ease, novel characters are referred to here by their item codes. There was greater corroboration between the categorisation and participant ratings of novel characters in general. However, there were areas of disagreement between the candidate's and participant's rating of the visual orientation and motor complexity of some characters. There was disagreement in the visual orientation of the characters NS_C and NS_I. The categorisation of NS_C was given a b-type orientation by the candidate whereas the predominant view amongst the participant raters was that it was d-type orientation. Similarly, the categorisation of NS_I was judged by the candidate to have a b-type orientation with three-strokes (3b) whereas the majority of participant raters judged the character to have a d-type orientation with two-stroke structure.

The remaining disagreements between original categorisation and the participant rating was in relation to the motoric complexity of the characters NS_G, NS_Q, and NS_U. The candidate categorised NS_G as three-strokes whereas the participant raters judged the character to be of two-strokes. On reflection, this character has a greater curvature and is graphically more distinct than the rest of the alphabet and novel characters and so the item was not selected for analysis. Both NS_Q and NS_U were categorised as having three-strokes each, whereas the participant ratings rated these characters as having two-strokes. Similarly, to the alphabet letters where there was a disagreement between the original categorisation and the raters judgement, the novel characters NS_Q and NS_U have curved aspects at the top and the bottom of the stem which is unlikely to have been treated as a separate stroke by the participant raters (see earlier section).

Table E.2.

Ratings of the Visual Orientation and Motor Complexity of Novel Characters by the Candidate and Participant Raters

Item	Code	Category	Participant Rating	% Participant Agreement						
				Orientation			Complexity			
				d	**n**	**b**	**1**	**2**	**3**	**4**
∋	NS_A	2d	2d	93	3	3	0	97	3	0
φ	NS_B	2n	2n	0	100	0	0	93	7	0
ɔ	NS_C	2b	2d	62	0	38	10	83	7	0
ч	NS_D	2d	2d	93	0	7	0	100	0	0
θ	NS_E	2n	2n	0	100	0	0	86	14	0
Ρ	NS_F	3b	3b	3	7	90	0	0	97	3
δ	NS_G	3d	2d	38	17	45	17	59	24	0
Ν	NS_H	3b	3b	3	0	97	0	7	93	0
Γ	NS_I	3b	2d	55	3	41	3	66	31	0
ⴹ	NS_J	3d	3d	86	3	10	0	7	93	0
Μ	NS_K	3b	3b	10	7	83	0	0	100	0
Ψ	NS_M	3n	3n	0	97	3	0	86	10	3
ꓵ	NS_N	2b	2b	31	21	48	0	97	3	0
o	NS_O	1n	1n	0	100	0	93	7	0	0
⋖	NS_P	3b	3b	21	7	73	0	24	76	0
ꓒ	NS_Q	3d	2d	97	0	3	0	62	38	0
Ͱ	NS_R	2b	2b	0	3	93	0	97	3	0
∞	NS_S	2d	2d	28	31	41	35	59	3	3
ⲧ	NS_T	3b	3b	41	3	55	0	7	90	3
ꓥ	NS_U	3d	2d	73	7	21	3	66	31	0
∠	NS_V	2b	2b	45	3	52	7	93	0	0
Њ	NS_W	4b	4b	0	7	93	0	3	0	97
∧	NS_X	2b	2b	0	100	0	3	97	0	0
ↄ	NS_Y	2d	2d	93	3	3	0	90	10	0
ⴺ	NS_Z	3d	3d	86	7	7	0	0	100	0

In sum, the agreement between original categorisation and the participant's ratings overall is good with excellent agreement on the visual orientation of the characters. However, there is disagreement in the motor complexity of characters amongst those which include a curve at the top or bottom of the stem. It appears participants have treated this curve as a continuation of the stem. However, the rationale for including these curves as separate stroke includes that the strokes distinguish between letters and curves reduce writing velocity.

Appendix F: Alphabet Letters and Novel Character Stimuli

Alphabet Letters			Novel Characters		
Item	No. of Segments	Orientation	Item	No. of Segments	Orientation
a	**2**	**d**	Э	2	d
b	**2**	**b**	φ	2	n
c	1	b	ꓛ	2	d
d	2	d	**Ч**	**2**	**d**
e	2	b	θ	2	n
f	**3**	**b**	**Ҏ**	**3**	**b**
g	**3**	**d**	δ	3	d
h	2	b	ᛈ	3	b
i	2	n	ᠴ	3	d
j	3	d	ᠴ	3	d
k	3	b	ᛘ	3	b
m	4	n	Ψ	3	n
n	**2**	**b**	ꞈ	2	b
o	1	n	o	1	n
p	2	b	⋖	3	b
q	3	d	ꝗ	3	d
r	2	b	Ⱶ	2	b
s	2	d	ᡐ	2	d
t	**3**	**b**	ᠴ	3	b
u	**2**	**d**	�	3	d
v	2	n	∠	2	b
w	4	n	Пɓ	4	b
x	2	n	∧	2	b
y	**3**	**d**	ᠪ	2	d
z	3	d	ᠵ	3	d

Note. Items selected for analysis are highlighted in bold.

Appendix G: Novel Character Visual Recognition Task

To ensure children had learned the visual form of novel characters and to examine whether this differed according to dyslexia or DCD we administered a short novel character recognition task immediately after training. Using E-prime 2.0 (Schneider, Eschman, & Zuccolotto, 2002), children were presented with either a novel character from the training (target) or a completely new novel character (distractor). They were asked to decide whether each character was one that they had written during the previous activity or not. The items appeared in the middle of the screen (Yuanti SC size 32 font) and remained on screen until a response was made. Children were instructed to respond as quickly as they could by pressing the 'z' (written today) or 'm' (not written today) keys. The presentation of items – 25 targets and 25 paired distractors – was randomised. Each distractor item was matched with a target item on 1:1 on both their visual orientation and the number of strokes.

Accuracy was high in all groups and the data was heavily skewed and so group medians were examined. Children with dyslexia (*Mdn* = 1) were more accurate than children with DCD (*Mdn* = .93), comorbid dyslexia and DCD (*Mdn* = .93), and typically developing children (*Mdn* = .93) at correctly identifying the target characters. A Kruskal-Wallis test confirmed there were no significant differences between groups in median accuracy of correctly identifying characters that had copied in the training session, $\chi^2(3) = 1.4$, $p = .706$. This findings suggests all children learned the visual form of the characters to a similar extent, regardless of group membership.