The Reciprocal Relationship between Oral Language and Reading Skills in Children with Down Syndrome

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Abstract

This thesis explored the interaction between oral language and reading skills in children with Down syndrome. Study 1 looked at the longitudinal relationship between reading accuracy, reading comprehension and oral language across three time-points. Study 2 taught new spoken words with or without the orthographic form present and Study 3 examined the effect of phonological pre-training on orthographic learning. In all three studies, typically developing children matched for reading accuracy also participated.

The effect of phonological and non-phonological oral language skills on reading accuracy was examined in Studies 1 and 3. In Study 1, phoneme awareness and vocabulary were evaluated as predictors of reading accuracy. Neither were longitudinal predictors in either group due to the strength of the autoregressor. However, vocabulary was a concurrent predictor for the children with Down syndrome. In Study 3, children with Down syndrome showed poorer orthographic learning than typically developing children. However when equated for decoding skill, the level of orthographic learning and the benefit from phonological pre-training were equivalent in the two groups.

The proposed benefit of learning to read on oral language development was tested in Studies 1 and 2. In Study 1, reading accuracy had a moderate effect on vocabulary development for children with Down syndrome and typically developing children. In Study 2, children with Down syndrome and typically developing children showed similar levels of phonological learning, which was facilitated by orthographic support to the same extent.

The relative contribution of reading accuracy and oral language to reading comprehension was evaluated in Study 1. Reading accuracy predicted concurrent reading comprehension for the children with Down syndrome and typically developing children, whereas oral language did not. However, only oral language was a longitudinal predictor in the children with Down syndrome. Theoretical and practical implications of the findings from all three studies are considered.

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Author's Declaration

I declare that the work presented within this thesis is my own work and has not been previously submitted for any other degree or qualification. Selected aspects of the research have been presented elsewhere:

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Chapter One

Reading Accuracy, Reading Comprehension and Oral Language Skills in Children with Down Syndrome

Overview

This thesis is concerned with the development of oral language and reading skills and how they interact in children with Down syndrome. This chapter will provide background information about Down syndrome and then focus specifically on predictors of reading accuracy and reading comprehension and also the impact of reading accuracy on oral language. Models of reading accuracy will be outlined to provide a theoretical framework in which to consider both the existing literature on children with Down syndrome and the findings of the empirical work within this thesis. The typical development of reading accuracy and reading comprehension, and the effect of reading accuracy on oral language in typically developing children will also be discussed in order to provide a basis for comparison with children with Down syndrome.

Introduction to Down Syndrome

Down syndrome is the most common genetic cause of learning difficulties and has a prevalence rate of 1.08 in every 1000 live births (Morris & Alberman, 2009). The most common form of Down syndrome is trisomy 21, which is caused by an extra copy of chromosome 21. Down syndrome can also be caused by mosaicism, when only some of the cells in the body are affected with trisomy 21, and Robertsonian translocation, when a section of chromosome 21 becomes attached to another chromosome during cell division. Down syndrome is typically associated with an IQ of approximately 50 although there is wide variation (Chapman & Hesketh, 2000; Määttä et al., 2011).

Social expectations have changed in more recent years and there has been a shift towards mainstream education, early intervention programmes and increased vocational training (Buckley, Bird, Sacks, & Archer, 2006; Chapman & Hesketh, 2000). As a group, individuals with Down syndrome make significant progress in literacy and numeracy whilst in education, although there is much individual variability, and a ‘plateau’ is often reported in early adulthood (Couzens, Cuskelly, & Haynes, 2011; Turner & Alborz, 2003).

Despite the variability, there appears to be a specific cognitive profile associated with Down syndrome. Verbal skills are typically a weakness compared to nonverbal skills (Abbeduto, Warren, & Conners, 2007; Jarrold, Baddeley, & Hewes, 1999). Within the oral language domain, expressive language is weaker than receptive language and there are greater impairments in grammar compared to vocabulary (Laws & Bishop, 2003). Relative strengths for individuals with Down syndrome include social skills, reading accuracy and visual memory (Abbeduto, 2008; Buckley, 1995; Fidler & Nadel, 2007; Jarrold et al., 1999; Klein & Mervis, 1999).

Individuals with Down syndrome often present with medical and physical complications including muscle hypotonia, abnormalities of the articulators, hearing problems and visual defects (Antonarakis, Lyle, Dermitzakis, Reymond, & Deutsch, 2004; Caputo, Wagner, Reynolds, Guo, & Goel 1989; Määttä et al., 2011; Marcell & Cohen, 1992; Schieve, Boulet, Boyle, Rasmussen, & Schendel, 2009; Shott, Joseph, & Heithaus, 2001). These have the potential to impact on learning and are therefore important to consider when studying cognitive development.

Individuals with Down syndrome have atypical speech-motor anatomy, which may then impact on speech development. Stoel-Gammon (1997) summarises the areas of difficulty as differences in vocal folds and oral cavity structure, weak facial muscles and hypotonicity affecting lip and tongue muscles. These problems may be expected to cause difficulties in acquiring normal speech patterns and more specifically, affect voice quality and articulation.

Hearing is often impaired in Down syndrome through abnormal hearing level thresholds and/or recurrent bouts of otitis media, a middle ear infection. Shott et al. (2001) found that 96% of children with Down syndrome had an ear infection during a five year period of observation, and of the children who required treatment for ear infections, 81% had abnormal hearing levels ranging from mild to severe impairments (see also Davies, 1996; Marcell & Cohen, 1992). If a child cannot hear language input adequately they may be expected to develop difficulties with both receptive and expressive language.

Visual defects are also common in Down syndrome, with a high incidence of astigmatism and refractive errors, e.g. short-sightedness and long-sightedness (Caputo et al., 1989). Without correction, this may affect a child's learning, particularly in regards to reading. However many visual problems are now easily corrected with spectacles, and therefore should not impact on learning significantly.

Studies of Reading Accuracy and Comprehension in Typical Development

This section aims to provide a backdrop for the studies conducted with children with Down syndrome, which will be discussed in a later section. Theoretical models of reading accuracy will be outlined and then key studies investigating longitudinal predictors of reading accuracy will be reviewed. Another aspect of literacy is reading comprehension, and this will be discussed in regards to the simple view of reading and the additional contribution of higher-level skills. In addition, the effect of reading accuracy on oral language development will be considered.

1.3.1. Reading Accuracy in Typical Development

Reading a word accurately can be achieved by two mechanisms: decoding and word recognition. Decoding occurs when the reader encounters an unfamiliar word and has to decipher it. This is often tested using nonword (or pseudoword) reading tasks. Word recognition, also referred to as word identification or sight word reading, refers to the ability to recognise and read words quickly and efficiently from memory and is crucial for skilled reading. In this thesis the term reading accuracy will be used to refer to tasks which require children to read words aloud, as using the term word recognition implies the singular use of that particular strategy whereas children could also be using decoding strategies.

Words differ in the regularity and consistency of their spelling-sound correspondences. Regular words are those which conform to grapheme-phoneme correspondences and therefore can be decoded. Irregular words, such as 'yacht', do not follow these grapheme-phoneme correspondences, and therefore cannot be decoded. A consistent word is pronounced like other words spelt in a similar way, for example the phonology of 'gave' follows the same pattern as 'save' and 'cave'. However inconsistent words do not follow this pattern, for example 'have' (Seidenberg, 2005).

1.3.1.1. Theoretical models of reading accuracy

1.3.1.1.1. Stage models.

1.3.1.1.1.1. Frith's stage model.

Marsh and colleagues (e.g. Marsh, Friedman, Welch, & Desberg, 1981) presented a stage model of reading accuracy where children move from rote learning to guessing from visual cues, to decoding and then to using analogies. Frith (1985) built on this to develop a model of reading accuracy, which consisted of logographic, alphabetic and orthographic stages. Children move through the stages in this fixed order and difficulty at one stage results in failure to move on to the next stage.

In the logographic phase, sight word reading dominates. Words are stored as whole units and children recognise them using visual cues. In this stage children acquire a large sight vocabulary but this is costly in terms of storage and can result in confusion between visually similar words. This prompts the beginning of the alphabetic stage. In this stage, children use their letter-sound knowledge and phonological skills to decode words in a serial manner. As children become proficient in both analytic skills and word recognition, they move into the orthographic stage where orthographic or morphological units within words are recognised.

Frith (1985) suggests that failure at different stages in the model can be used to explain impairments in reading accuracy. Failure at the logographic stage results in an inability to acquire a sight vocabulary; however the existence of some, if limited, reading ability in children with developmental delays suggests that this is rare in practice. Difficulty at the alphabetic stage is more common and results in problems in reading and spelling unknown words as seen in dyslexia. Although children may be able to compensate to some extent, this does not address the underlying problems in phonological skills and due to the sequential order of the stages the child cannot move on to the orthographic stage. Failure at the orthographic stage results in difficulty spelling irregular words, however due to intact logographic and alphabetic strategies children are able to read irregular words and read and spell unknown regular words.

1.3.1.1.1.2. Ehri's phase model.

Ehri (e.g. Ehri, 2002; Ehri, 2005a; Ehri, 2005b; Ehri & McCormick, 1998) proposed a phase model of sight word reading development. This model of sight word reading is emphasised to consist of *phases* rather than stages, which carries the implication that there are less distinct divides between the four periods of development: the pre-alphabetic, partial alphabetic, full alphabetic and consolidated alphabetic phases.

The pre-alphabetic phase is characterised by the use of visual cues, within the word or surrounding environmental print including logos, and the absence of letter knowledge. In this stage, sight word reading is achieved by associating the visual properties of the word with its meaning. No phonological information is involved; rather it is argued that the pronunciation of the word is activated after the semantic representation.

The partial alphabetic phase begins with the emergence of letter-sound knowledge and phoneme awareness. Children can use this knowledge to form partial connections between spellings and pronunciations, often relying on initial or final letters. Ehri (2005a) gives the example of a child knowing the phonemes for the graphemes 'j' and 'l' and therefore making a successful attempt at reading 'jail'. Hence sight word memory is now beginning to be alphabetic, i.e. phonological information is connected to the orthographic representation. With increasing letter-sound knowledge and phoneme awareness, children progress into the full alphabetic phase. They are now able to fully decode unfamiliar words. This allows words to be securely stored in memory, with complete rather than partial phonological information. The final stage, the consolidated alphabetic phase, occurs when children begin to use morphological and orthographic units to read unfamiliar words.

Within this model, children with reading difficulties, specifically dyslexia, may be using mostly partial-alphabetic strategies (Ehri & McCormick, 1998; Ehri & Saltmarsh, 1995). Children have incomplete knowledge of letter-sound correspondences and/or phonemic awareness, and therefore find it difficult to apply fully analytic strategies to words. As a result, their sight word vocabulary is slower to develop.

1.3.1.1.1.3. Summary: Stage models of reading.

Stage, or phase, models of reading accuracy are explicitly developmental, describing in detail behaviour and possible errors at each stage. The term 'stage' suggests that success at each stage is necessary to progress to the next one and that each stage is qualitatively unique. However Ehri (e.g. 2005b) emphasises the use of the term 'phase' to highlight that not each phase is a prerequisite for the next, e.g. recognising environmental print may not actually help a children progress to the partial alphabetic phase, and children may use different strategies within the same phase. Although stage models provide rich descriptions of behaviour at given points in the development of reading accuracy, they do not offer causal explanations of the underlying mechanisms. Furthermore individual differences are neglected (Hulme & Snowling, 2009).

1.3.1.1.2. Computational models of reading accuracy.

Computational models are computer simulations of a process or behaviour, for example reading. There are several advantages of using computational models to study reading (Coltheart, 2005). Importantly the theory behind such simulations has to be explicit and well-specified in order to be implemented in the model. Furthermore the model's behaviour can be compared against human behaviour. Here the two most influential computational models of reading accuracy, the dual route model and the triangle model, will be discussed.

1.3.1.1.2.1. Dual route model.

The dual route model of reading proposes a lexical and a non-lexical route to reading aloud (e.g. Coltheart, 2005; Coltheart, Curtis, Atkins, & Haller, 1993). The lexical route involves direct retrieval of words already stored in the lexicon and the non-lexical route uses grapheme-phoneme correspondences to read words. The lexical route is used to retrieve familiar words, whereas nonwords have to be read by the non-lexical route. The non-lexical route can also successfully read regular familiar words but as a result of using grapheme-phoneme correspondences 'regularises' irregular words. The dual route cascaded (DRC) model is a computational model representing a skilled adult reader. It has been able to replicate many effects found in skilled readers and reads words and nonwords with a high level of accuracy (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001).

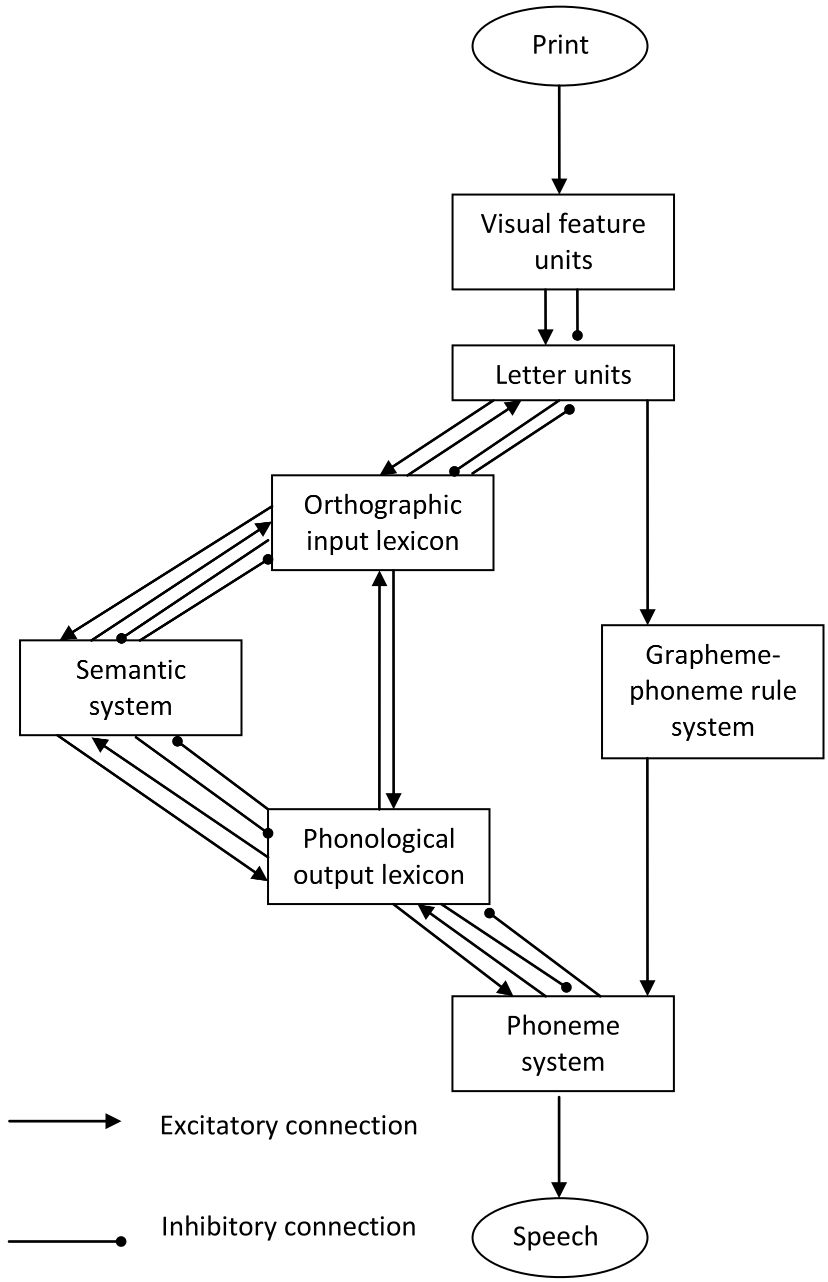


Figure 1. The DRC model of reading e.g. Coltheart et al. (2001)

As can be seen in Figure 1 there are three routes depicted for turning print into speech: the non-lexical route, the lexical non-semantic route and the lexical semantic route, although the latter has not yet been implemented (for an implementation of a minimal semantic system see Coltheart, Woollams, Kinoshita, & Perry, 1999). The DRC model utilises localist representations, i.e. words are represented as whole units. In the lexical route the visual features of letters activate letter units, which activate the word unit in the orthographic lexicon. This in turn activates the corresponding entry in the phonological lexicon, which activates the individual phonemes. The non-lexical route bypasses the lexicon, applying grapheme-phoneme correspondence rules to the input letters individually from left to right. The excitatory and inhibitory connections between the different units mean that semantic information could also be activated by the non-lexical route and lexical non-semantic route. Any semantic activation would strengthen the activation for the corresponding orthographic representation, thus presumably supporting the search of the lexicon (McKague, Pratt, & Johnston, 2001). The routes within the DRC model operate simultaneously and therefore conflicts can arise over the pronunciation of nonwords, which would be read correctly by the non-lexical route, and irregular words, which would be read correctly by the lexical non-semantic route.

1.3.1.1.2.2. Triangle model.

Connectionist models attempt to represent processing in the brain by implementing neuron-like units and connections. The 'triangle' connectionist model of reading aloud stands in contrast to the DRC model as it conceptualises reading as a statistical learning problem, therefore it is able to speak to reading accuracy development. There have been several versions of the triangle model since it was first proposed by Seidenberg and McClelland (1989) but all have the basic structure of connections between phonology, semantics and orthography (see Figure 2). Phonology can be activated from orthography in two ways: directly or indirectly via semantics. This distinction between the two pathways is similar to that in the DRC model. However there are important differences in the architecture of, and processes within, the models.

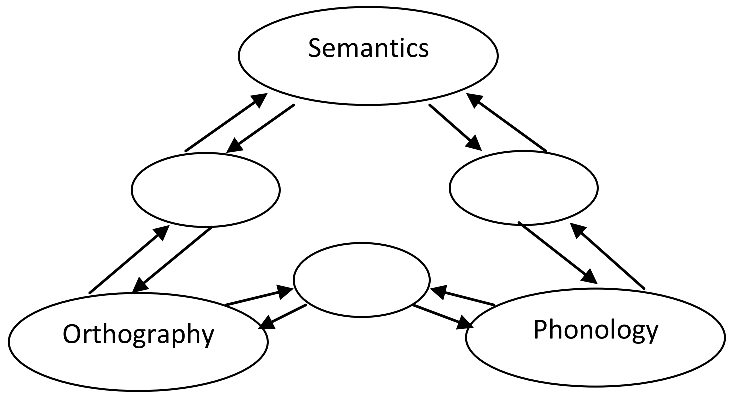


Figure 2. The triangle model of reading e.g. Plaut et al. (1996)

Rather than the localist representations used by the DRC model, the triangle model utilises distributed representations, i.e. words are represented as patterns distributed across units. There are separate groups of units that represent components of phonology, orthography and semantics, for example visual features of letters, the letter's position within a word and phonetic features of the phoneme. Between these sets of units are hidden units which permit learning. When undertaking the task of reading a word aloud, the pattern of activation at the orthographic level flows through the hidden units, either directly or via the semantic units, and produces a specific pattern of activation at the phonological level. The network learns by comparing this generated output against the correct output and altering the weights, or connections, within the model accordingly. Therefore words are represented by a particular pattern of activity, rather than storing whole words in the lexicon.

The triangle model was first proposed by Seidenberg and McClelland (1989). The model was highly successful at reading trained words and replicated the effects of frequency (i.e. high frequency words being read quicker than low frequency words) and consistency (i.e. consistent words being read quicker than inconsistent words) found in human readers. However, the model was poor at pronouncing nonwords. In this model, the orthographic input was coded as context-sensitive letter triples and phonological output was coded as triples of phonemic features. Plaut, McClelland, Seidenberg and Patterson (1996) improved upon this by basing orthographic and phonological representations on individual graphemes and phonemes. As a result the model's nonword reading reached the same standard as human readers. In a further simulation, the semantic pathway was lesioned and performance worsened on exception words, particularly those of low frequency. However semantics was conceptualised as an additional input to the phonological representation, rather than information about the meaning of the word.

1.3.1.1.2.3. Summary: Computational models of reading.

Both the DRC and triangle models have successfully simulated aspects of human reading behaviour (e.g. Coltheart et al., 2001; Harm & Seidenberg, 2004; Plaut et al., 1996). Importantly for the purposes of this thesis, the DRC model is not developmental as its architecture is pre-specified and represents an already skilled reader. In contrast, the triangle model learns through the re-adjustment of weights in response to feedback; although it should be highlighted that the presence of explicit feedback for every attempt at a word does not reflect the typical experience of a child.

1.3.1.2. Phonological awareness, letter-sound knowledge and the alphabetic principle.

Phonological awareness is considered to be "the ability to reflect explicitly on the sound structure of spoken words" (Hatcher, Hulme, & Ellis, 1994, p. 41), and includes large units such as syllables and rimes, and smaller units such as phonemes. Tasks used to measure phonological awareness range from the relatively simple, for example where children hear several words and choose which one starts with a target sound, to more complex tasks, for example where children have to delete phonemes from words.

The alphabetic principle is the knowledge that graphemes represent phonemes, and that a particular phoneme is represented by the same grapheme regardless of the position it occurs in a word (Byrne & Fielding-Barnsley, 1989). In a seminal study which demonstrated the importance of the alphabetic principle for decoding, Byrne and Fielding-Barnsley taught children a graphemic representation for two words such as 'sat' and 'mat' using symbols to represent onsets and rimes. They were then tested on transfer items in a forced-choice task, which could be distinguished by recognition of the previously learnt symbols. Children who were successful on both phoneme awareness and symbol-phoneme correspondence tasks during training were more likely to pass the transfer test. Byrne and Fielding-Barnsley suggested that explicit instruction in grapheme-phoneme correspondences along with the knowledge that words are composed of smaller speech segments, i.e. phonemes, is necessary for the acquisition of the alphabetic principle. However they note that other processes, namely assembling the identified phonemes into a whole word pronunciation, are also required for successful decoding.

There is a wealth of longitudinal studies highlighting the important role that both phonological awareness and letter-sound knowledge have in the development of reading accuracy skills. Wagner et al. (1997) found that phonological awareness accounted for variance in later reading accuracy independently of earlier reading accuracy and this relationship continued throughout children's early schooling from age five to ten years. Muter, Hulme, Snowling and Stevenson (2004) conducted a two-year longitudinal study with children who had just started school in the UK, hence were aged 4-5 years. They found that both phoneme awareness and letter knowledge predicted later reading accuracy, after controlling for the autoregressor. The finding that phonological awareness and letter-sound knowledge are strong predictors of reading accuracy is robust and has been replicated extensively (e.g. Bryant, MacLean, Bradley, & Crossland, 1990; Muter, 1994; Stuart & Masterson, 1992). It is important to note here that although Muter et al.'s longitudinal study found that phoneme awareness was highly predictive of later reading accuracy, rime awareness was not. This suggests that there is a division within phonological awareness between rimes and phonemes, with the latter being more important for reading accuracy development.

The proposed causal role of phonological awareness and letter-sound knowledge in reading accuracy has been strengthened by training studies. An influential study by Bradley and Bryant (1983) found that children who were trained in sound categorisation and letter-sound knowledge achieved higher scores on reading accuracy tasks than children who received no intervention and a group of children who received semantic categorisation training to act as a control condition. Other studies have also shown that training phonological awareness and letter-sound knowledge consistently leads to improvements in reading accuracy and spelling (e.g., Fox & Routh, 1984; Lundberg, Frost, & Peterson, 1988; Schneider, Küsbert, Roth, Visé & Marx, 1997; Schneider, Roth & Ennemoser, 2000; Treiman & Baron, 1983).

Hatcher et al. (1994) formally proposed that training that explicitly links phonological skills and reading accuracy would be more effective than training phonological skills alone; this was termed the 'phonological linkage hypothesis'. To test this, the relative effectiveness of training phonological skills alone, reading accuracy alone or phonological and reading accuracy skills combined was compared to an untreated control group. In the combined programme the links between phonology and reading accuracy were emphasised using activities largely based on letter-sound knowledge. Only the group who received the combined programme scored significantly above the control group on immediate and delayed tests of reading accuracy. These results offer a clear message: phonological awareness needs to be taught in the presence of print, and crucially these two skills need to be explicitly linked with reference to letter-sound knowledge, to produce maximum gains in reading accuracy.

1.3.1.3. Non-phonological oral language skills.

Oral language consists of several separable domains: phonology (sounds within words), semantics (meanings of words), pragmatics (social use of language) and grammar which can be subdivided into morphology (units of meaning) and syntax (structure of phrases and sentences). Therefore beyond phonological skills, there are broader oral language skills that, although have received less attention than phonological skills in relation to reading accuracy, appear to contribute to development of this skill.

The lexical restructuring hypothesis states that as a child's spoken vocabulary increases in size, phonological representations become more fine-grained (e.g. Walley, Metsala, & Garlock, 2003). In early vocabulary development, words can be stored holistically but as the number of spoken words in the lexicon increases, phonological representations need to become better specified in order to remain distinct. Therefore an indirect relationship between oral vocabulary and reading accuracy via development of phonological awareness is posited. Storch and Whitehurst (2002) examined the direct and indirect relationship between oral language and reading accuracy by conducting a longitudinal study with 626 children in the USA. Their oral language and literacy skills were assessed prior to age four to starting school and then every school year until grade 4 (9-10 years). Reading accuracy in grades 1-4 was predicted by earlier phonological awareness and letter knowledge, and oral language had an indirect effect on reading accuracy which was mediated by these skills. Lonigan, Burgess and Anthony (2000) similarly found that oral language predicted later phonological awareness and letter knowledge in preschool children. Therefore oral language may promote the early development of the skills necessary to acquire the alphabetic principle so children can become successful readers.

In contrast, a study with 1137 children by NICHD Early Child Care Research Network (2005) found that oral language at age 4;06 had a direct effect on reading accuracy at age 6-7 years. There was also an indirect pathway to reading accuracy via phonological awareness. There is a difference in the phonological awareness tasks used in this study and Storch and Whitehurst (2002) that could account for the difference in results. The phonological awareness task in this study required children to listen to an incomplete word, e.g. ba..tub, and produce the intended word, e.g. bathtub. Although the authors argue that the child must be sensitive to and manipulate phonemes in this task, there is clearly a large vocabulary component as if children did not know the word ‘bathtub’ they would be unable to complete this task. Indeed other studies, along with Storch and Whitehurst, with purer tests of phoneme awareness have found that oral language does not add unique variance to reading accuracy (Muter et al., 2004) but does contribute to phonological awareness (Sénéchal, Ouellette, & Rodney, 2006), and therefore the weight of evidence argues against a direct relationship between oral language and reading accuracy in early literacy development.

In later childhood, it may be that the effects of phonological awareness and letter-sound knowledge on reading accuracy become less important and therefore a direct link between oral language and reading accuracy may be more evident. In a longitudinal study Nation and Snowling (2004) examined the effect of broader oral language skills on reading accuracy development between the ages of eight and 13 years. Assessments of vocabulary definitions, listening comprehension and a composite measure of semantics administered at the first time point all contributed uniquely to later reading accuracy, even after initial reading accuracy and phonological skills were controlled for. This relationship may occur because children with good oral language skills may develop larger sight word vocabularies because they are independently able to use context to acquire new words. Considering the triangle model, another possible explanation is that the combination of activation from *both* semantic and phonological pathways results in faster and more accurate word recognition.

Nation and Cocksey (2009) drew a distinction within vocabulary tasks between lexical phonology (familiarity with the phonological form of a word) and semantics (deeper knowledge about a word's meaning). In their study lexical phonology was tested using a lexical decision task. Children were also asked to define the words, to measure semantic knowledge. At an item-level, it was the measure of lexical phonology which uniquely contributed to whether children could read the words, and there was no additional effect from their semantic knowledge of the words. The words used were irregular, and the authors argue that familiarity with the phonological form of the word allows children to produce the correct answer on the basis of a partial decoding attempt.

A distinction within vocabulary between breadth and depth of knowledge has also been made (Ouellette, 2006). Breadth refers to the number of lexical entries a child may have and vocabulary depth refers to the semantic information a child possesses about a particular word. To assess the independent contributions that these components of vocabulary may make towards reading accuracy, 60 children aged 9-10 years were administered tasks of receptive vocabulary breadth, expressive vocabulary breadth, expressive vocabulary depth, nonword reading and irregular word reading to measure word recognition. Vocabulary breadth, as assessed by a receptive multiple-choice task, accounted for unique variance in nonword reading, which Ouellette suggests is due to the importance of detailed phonological representations in both nonword reading and for the existence of a large lexicon. Irregular word reading was predicted by both breadth and depth, but only expressive vocabulary breadth accounted for independent variance. The expressive vocabulary breadth task was a picture naming task and Ouellette argued that the procedure of encoding, organising and retrieving words is what may, at least, partly drive the correlation between picture naming and word recognition.

Morphology and its relationship to reading accuracy has also been investigated. Roman, Kirby, Parrila, Wade-Woolley and Deacon (2009) conducted a concurrent study with children aged 9-14 years and found that morphological awareness predicted reading accuracy. However, a vocabulary task was not included in this study and therefore it is not known whether the contribution of morphology is shared with vocabulary. In a different study with children of a similar age, morphological awareness was found to contribute uniquely to measures of spelling and reading accuracy after vocabulary had been accounted for (Nagy, Berninger, & Abbott, 2006).

Different types of words can be included in single word reading tasks, for example exception words, regular words and nonwords. Ricketts, Nation and Bishop (2007) investigated how vocabulary may contribute to separate reading tasks for each of these categories of words. It was found that vocabulary, as assessed with a definitions task, was a unique predictor of exception word reading only. This relationship was suggested to arise because exception words cannot be decoded; therefore whether the word is already stored in a child's lexicon becomes more important.

1.3.1.4. Memory

Working memory is considered to be a system with limited capacity that temporarily stores and processes information. According to Baddeley and Hitch's model (Baddeley, 2000; Baddeley & Hitch, 1974), the working memory system consists of the phonological loop, visuospatial sketchpad, central executive, and the episodic buffer. The phonological loop is for the temporary storage of phonological information, which decays unless refreshed by subvocal rehearsal and the visuospatial sketchpad is a short-term store for visual and spatial information. The central executive is a domain-general attentional control system and the episodic buffer is a system that allows information from different sources to be integrated.

Different tasks are used to measure different aspects of working memory. Tasks that require the storage of verbal information, for example remembering lists of spoken words or digits which increase in length, are thought to rely on the phonological loop. Similarly tasks which require the storage of visuospatial information, for example the Corsi blocks task that requires children to remember a sequence of locations on a board, tap the visuospatial sketchpad. Tasks which require simultaneous storage and processing of information, for example recalling a list of digits backwards, are proposed to recruit the central executive along with the modality-specific short-term store.

Nevo and Breznitz (2011) conducted a one-year longitudinal study to examine the contribution of memory skills to early reading development in Hebrew. It was found that visuospatial short-term memory accounted for unique variance in later reading speed. Other studies (e.g. McDougall, Hulme, Ellis, & Monk, 1994) have not found a link between visual memory and reading accuracy, and it may be that this effect is specific to Hebrew. Verbal working memory was found to predict unique variance in later reading accuracy and speed. Importantly this was unique from IQ and phonological awareness, which is in contrast to other studies (e.g. Durand, Hulme, Larkin, & Snowling, 2005).

In Nevo and Breznitz's (2011) study reading accuracy was not assessed at the first time point and therefore the autoregressive effect cannot be controlled for and this is the most stringent test of whether there is a longitudinal predictive relationship between two skills. Indeed other studies have found that verbal memory does not predict later reading accuracy after controlling for the autoregressor and initial phonological awareness (Lervåg, Bråten, & Hulme 2009; Wagner et al., 1997). Furthermore speech rate was not accounted for and previously it has been found that when speech rate is controlled for, verbal memory does not account for unique variance in reading accuracy (McDougall et al., 1997).

The phonological loop is thought to be important for learning new words, and its primary role is to generate representations of new words rather than repeat digit or word sequences (Baddeley, Gathercole, & Papagno, 1998). Therefore the task of nonword repetition has been employed to assess the phonological loop. This task and what underlying skills it measures has been the subject of intense debate. Initially it was proposed that nonwords do not activate existing lexical representations and therefore nonword repetition is a purer test of phonological storage than word or digit recall; however it appears that this is too simplistic a picture and linguistic knowledge also affects performance on this task (Gathercole, 2006; Gathercole & Baddeley, 1989; Gathercole, Willis, Emslie, & Baddeley, 1991; Snowling, Chiat, & Hulme, 1991). Indeed it could be argued that nonword repetition is primarily a phonological task, involving segmentation of the phonological input, generating speech motor programs and articulating the phonological output (Snowling et al., 1991).

Muter and Snowling (1998) assessed the contribution of nonword repetition to word and nonword reading. Children were assessed at age four, six and nine years on a range of measures including IQ, reading accuracy, nonword reading, nonword repetition and phoneme awareness. After IQ was controlled for, there was a significant effect of nonword repetition at age six and nine years on later word and nonword reading but not from when children were aged four years. However phoneme awareness was not controlled for and as phoneme awareness and nonword repetition were moderately correlated, nonword repetition may not be a unique predictor of reading accuracy and/or decoding beyond phoneme awareness.

Nation and Hulme (2011) assessed the reciprocal relationship between nonword repetition and reading accuracy at two time-points when children were aged 5-6 years and 6-7 years, whilst controlling for phoneme deletion, oral language and the autoregressors. The longitudinal relationship from earlier nonword repetition to later reading accuracy was not significant. However reading accuracy predicted later nonword repetition suggesting nonword repetition is a consequence of reading accuracy rather than a cause, a point which will be discussed in more detail in a later section.

1.3.1.5. Summary: Reading accuracy in typical development.

There is an abundance of evidence to support the importance of phonological awareness, specifically at the phoneme level, and letter-sound knowledge in learning to read. These skills are important because together they enable children to grasp the alphabetic principle. Many of these studies have been conducted with beginning readers and therefore highlight the importance of phoneme awareness and letter-sound knowledge at this early stage of learning to read. However once children have grasped the alphabetic principle and are able to decode accurately, then the development of phoneme awareness and letter-sound knowledge is likely to asymptote and therefore other skills may contribute more to reading accuracy development.

Beyond phonological skills, oral language may have both direct and indirect effects on reading accuracy. In the early stages of learning to read oral language, particularly vocabulary, is argued to play a role in the development of phonological awareness, and therefore indirectly promotes reading accuracy. There is also some evidence for a direct relationship between oral language and reading accuracy in later development, particularly when children are employing word recognition strategies rather than decoding. However recent evidence suggests this may be due to lexical phonology rather than semantic knowledge per se when considering the effect of vocabulary (Nation & Cocksey, 2009). This is a relatively new direction of research, and it will be crucial to see whether this finding is replicated.

As reading is a verbal task, it is unsurprising that the relationship with verbal memory measures tends to be higher than with visual memory. There is some evidence that verbal working memory may be related to reading accuracy, and indeed this seems intuitive if considering the memory load of decoding an unfamiliar word. However it appears that this relationship may reflect other skills including IQ, phonological awareness and speech rate. Furthermore nonword repetition appears not to predict reading accuracy, at least in the early stages of reading development.

1.3.2. Reading Comprehension in Typical Development

1.3.2.1. The simple view of reading: The role of reading accuracy and oral language.

The ultimate goal of reading is to comprehend text. Clearly, being able to read words accurately is important to achieve this goal, but so is language comprehension. Gough and Tunmer (1986) proposed the simple view of reading, a model which states that reading comprehension is the product of both decoding and linguistic comprehension. Therefore a child needs adequate levels of both skills to understand text.

The relative roles of reading accuracy and oral language have been suggested to differ throughout development. In the convergent skills model of reading development Vellutino, Tunmer, Jaccard and Chen (2007) suggest that a sufficient level of reading accuracy must be reached so children can access the same level of written language as in the spoken domain. When this occurs, language comprehension then plays the dominant role in reading comprehension. In support of this, the pathway between reading accuracy and reading comprehension was significant in a group of younger readers (aged 7-9 years) but not in a group of older readers (aged 11-13 years). Although significant in both groups, the pathway between language comprehension and reading comprehension was significantly stronger in the older than younger children. Similarly Ouellette and Beers (2010) found that the contribution of reading accuracy to reading comprehension was higher in a group of children aged 5-7 years than a group of children aged 11-12 years, and conversely the contribution of oral language was higher in the older group than younger group. These results suggest that with age, oral language becomes more important and reading accuracy becomes less important in predicting reading comprehension.

Ouellette and Beers (2010) also examined the contribution of different facets of reading ability and oral language to reading comprehension. Decoding and word recognition were assessed with tasks of nonword reading and irregular word reading. In the younger group of children, both nonword and irregular word reading contributed unique variance to reading comprehension. In the older group of children, neither contributed unique variance. However phonological awareness did, and as this was entered first in a hierarchical regression it may have subsumed some of the same variance as in the reading measures, particularly nonword reading.

To investigate the relationship between different aspects of spoken language and reading comprehension, Ouellette and Beers (2010) used tasks of listening comprehension, vocabulary breadth and vocabulary depth. For children aged 5-7 years, no language measure contributed unique variance to reading comprehension after word and nonword reading measures were entered. For children aged 11-12 years, vocabulary breadth accounted for unique variance after the effects of word and nonword reading and listening comprehension were controlled for. Therefore later in development when the effect of language appears to be more important, vocabulary appears to have a specific role on reading comprehension beyond that of general understanding of language. In support of this, the relationship between vocabulary breadth and reading comprehension has also been found to be significant in other cross-sectional studies (Storch &Whitehurst, 2002). Ouellette and Beers did not assess grammar, but as understanding a text also requires comprehension at the sentence level, it is likely that tasks of sentence comprehension may also be related to reading comprehension. Indeed such a relationship has been found to be significant in other cross-sectional studies with children aged 7-15 years (Nagy, Berninger, Abbott, Vaughan, & Vermeulen 2003; Nagy et al., 2006).

There is good evidence for a concurrent relationship between reading comprehension and vocabulary. However it may be that higher levels of reading comprehension result in children being able to acquire the meanings of new words from surrounding context. Therefore longitudinal studies and intervention studies are needed to elucidate the direction of this relationship. In early literacy development, Muter et al. (2004) found that earlier measures of both vocabulary and grammar accounted for unique variance in reading comprehension at age 6-7 years. The NICHD Early Child Care Research Network (2005) found that reading comprehension at age 8-9 years was predicted by earlier measures of general receptive and expressive language and a picture naming task. Similarly Oakhill, Cain and Bryant (1997) found that vocabulary, but not grammar, predicted later reading comprehension when children were aged 8-9 years. However none of these studies accounted for earlier reading comprehension skill, possibly as reliable measures of reading comprehension can be difficult to obtain in early literacy development as children need to have reached a sufficient level of reading accuracy. Nation and Snowling (2004) investigated the predictors of reading comprehension over and above the autoregressive effect in later childhood. They found that listening comprehension and vocabulary when children were aged 7-10 years predicted reading comprehension when children were aged 12-14 years, after controlling for both the autoregressor and decoding ability.

Children known as poor comprehenders have age-appropriate reading accuracy but weaknesses in reading comprehension. They also present with oral language weaknesses (Nation, Clarke, Marshall, & Durand, 2004). Comparisons have been drawn between poor comprehenders and children with Down syndrome, which will be discussed later in this chapter. Clarke, Snowling, Truelove and Hulme (2010) conducted a randomised controlled trial with children with poor reading comprehension, which compared three intervention programmes compared to a waiting control condition. One programme trained oral language (e.g. vocabulary, spoken narrative), one trained text comprehension skills (e.g. comprehension strategies, written narrative) and another combined these two approaches. All groups made significant gains compared to a waiting control group but the oral language programme resulted in the largest gains at follow-up and this was partially mediated by vocabulary skill. This provides converging evidence to support an important role for oral language in reading comprehension skill, with a particular emphasis on vocabulary.

1.3.2.2. Higher-level processing.

Kintsch and Rawson (2005) suggest that there are higher-level processes involved in reading comprehension beyond reading accuracy and lower level language skills. They propose that individuals must understand both the microstructure and the macrostructure of a text. The microstructure refers to understanding word meanings and how they group together into idea units, or propositions. This local representation of the text is in contrast to the macrostructure, or the global structure, which is concerned with how topics within the text relate to each other. The microstructure and macrostructure combine to form the textbase. This, along with the reader’s prior knowledge, results in a situation model, or a mental model of the text. To create a situation model, processes such as working memory, inferencing, comprehension monitoring and knowledge of story structure are required.

Reading comprehension requires the processing of information and, in the context of Kintsch and Rawson’s (2005) theory, holding this information in memory to create a situation model. Therefore working memory is necessary for successful reading comprehension. Seigneuric, Ehrlich, Oakhill and Yuill (2000) looked at the contribution of verbal, numerical and spatial working memory to reading comprehension in a group of 48 children aged 8-10 years. Regression analyses were conducted to investigate whether working memory contributed unique variance to reading comprehension when decoding and vocabulary were also entered. The verbal working memory tasks were significant predictors whereas the other tasks were not. This supports Kintsch and Rawson, particularly as the verbal domain would be the most likely to support reading comprehension through the storage and integration of information gathered from the text.

Sesma, Mahone, Levine, Eason and Cutting (2009) examined the contribution of memory and executive functioning to reading comprehension in a group of 60 children aged 9-

15 years; half of whom had difficulties in reading accuracy, reading comprehension and/or attention. After decoding, vocabulary and parental reports of attention were controlled for, measures of working memory and planning contributed unique variance to reading comprehension. However this measure of working memory included arithmetic and digit span therefore was not a pure task.

If working memory contributes to reading comprehension, then children with specific reading comprehension difficulties, i.e. poor comprehenders, might be expected to have weaknesses in working memory. Indeed a meta-analysis by Carretti, Borella, Cornoldi and De Beni (2009) found that poor comprehenders showed difficulties on short-term and working memory tasks, although verbal working memory tasks posed the most difficulty for this group. They also had weaknesses on tasks of executive functioning, which required inhibiting irrelevant information and updating information in memory.

Some higher-level processes proposed to be involved in reading comprehension are more specific to this task, including inference-making, comprehension monitoring and story structure knowledge. When reading a text, it is often necessary to go beyond the information explicitly given in the text and make an inference, for example understanding who a pronoun refers to (cohesive inferences) or the application of real-world knowledge (elaborative inferences). Furthermore in order to fully understand text, the process of comprehension monitoring must occur; this enables individuals to be aware of when their understanding breaks down and attempt to address this (Perfetti, Landi, & Oakhill, 2005). Knowledge of story structure is also important. This is the production and understanding of different components and the sequence of a narrative, for example titles, the introduction of characters and endings (Perfetti et al., 2005).

As with working memory, the level of inferencing, comprehension monitoring and story structure have been examined in poor comprehenders. Cain, Oakhill, Barnes and Bryant (2001) taught children information about an alien planet to ensure that any differences in inference-making were not due to differences in existing knowledge. Compared to skilled comprehenders who were matched for reading accuracy and age, poor comprehenders made less successful inferences, and this difference remained when performance on literal questions (those which did not require an inference) and retention of taught knowledge were controlled for. The authors suggested this result arises because poor comprehenders have difficulty in selecting the appropriate piece of information for making the inference.

A difference between skilled and poor comprehenders has also been found in resolving anomalies within text, knowledge of story conventions and structure of stories produced (Cain, 1996; Yuill, Oakhill, & Parkin, 1989). In the latter case, poor comprehenders also told poorer structured stories than younger children matched on comprehension skill when given only a general topic as a prompt. When children were provided with a sequence of stories, the poor comprehenders and the comprehension-matched group told comparable stories (Cain, 1996). It was suggested that the poor comprehenders were less able to independently produce a structural framework for a cohesive story.

Oakhill et al. (2003) conducted a longitudinal study of reading comprehension and included measures of vocabulary, working memory, comprehension monitoring, inference comprehension, narrative sequencing and story structure knowledge. Ninety-six children participated in two time points, when they were aged 7-8 years and 8-9 years. Verbal working memory, comprehension monitoring, inference comprehension and story structure knowledge all predicted unique variance in reading comprehension after vocabulary skills were accounted for. The most stringent test would be to control for the autoregressive effect of earlier reading comprehension, which was not done here. Furthermore reading accuracy was not entered as a control variable, so the effect of these variables beyond the contribution of reading accuracy cannot be determined.

1.3.2.3. Summary: Reading comprehension in typical development.

Reading comprehension was originally envisaged as a product of both decoding and oral language comprehension. At the most basic level, this appears to be true but this fails to capture changes across development, the separable components within decoding and oral language comprehension and other higher level processes such as working memory. In the early stages of literacy development, reading accuracy constrains reading comprehension and therefore is a stronger predictor than oral language. As children's reading accuracy develops, the role of oral language as a predictor increases. There are many different skills within oral language and vocabulary appears to be particularly important. Along with reading accuracy and oral language, higher-level processes such as working memory, inferencing, comprehension monitoring and story structure knowledge also contribute to reading comprehension skill, although it may be these can be reduced to a more general oral language factor (Hulme & Snowling, 2011).

1.3.3. Impact of Reading Accuracy Development on Oral Language

The effect of oral language, particularly phonological skills, on reading accuracy has been extensively studied as discussed above. In contrast, there has been much less research on the reverse relationship, i.e. the impact of reading accuracy on oral language. This will be discussed in regards to children with Down syndrome later in this chapter, and therefore studies in typical development are relevant.

Ziegler, Muneaux and Grainger (2003) considered the effect of orthography on phonology in the context of the lexical restructuring hypothesis; they argued that as the number of acquired orthographic representations increases, this may also promote segmental phonological representations. This can be related to Perfetti and Hart's (2002) lexical quality hypothesis, of which the key premise is that a lexical representation is high quality when it has fully specified orthographic, phonological and semantic representations. It is also suggested that reading practice can produce better specified lexical representations and one mechanism by which this occurs could be by promoting the segmental nature of phonological representations.

A number of studies have taken advantage of an opportunity to avoid the confounds of social or medical factors to study the effect of illiteracy in a Portuguese community. These studies have focused on phonological skills and have found that adults who never learnt to read were poorer on phonological awareness tasks and nonword repetition than those who had learnt to read (Castro-Caldas, Petersson, Reis, Stone-Elander, & Ingvar 1998; Morais, Cary, Alegria, & Bertelson, 1979). Morais et al. (1979) suggested that the explicit awareness and manipulation of phonemes is not a necessary prerequisite for learning to read, but the underlying cognitive ability for this awareness to develop is, and there is a reciprocal relationship between development in reading accuracy and phonological awareness.

Hulme, Caravolas, Malkova and Brigstocke (2005) outline two versions of the theory that increasing orthographic knowledge promotes phonological awareness. The strong version is that there is an item-specific relationship between acquiring grapheme-phoneme correspondences and phoneme awareness, i.e. children learn to successfully manipulate phonemes they know the letter for. A second, weaker, account of the relationship is that learning grapheme-phoneme correspondences results in children acquiring the alphabetic principle and the knowledge that spoken words can be segmented into their constituent phonemes. However in this study, children successfully completed phoneme awareness tasks for phonemes they did not know the letter for and some children with no letter-sound knowledge succeeded on phoneme awareness tasks. This contradicts both versions of the theory, particularly the stronger one. Hulme et al. conclude that although there may be a reciprocal relationship between orthographic and phonological relationships, learning to read is not a necessary prerequisite for phoneme awareness to develop.

If learning to read does indeed affect oral language skills, then it may be expected that this would lead to changes in the functional organisation of the brain. To investigate this Castro-Caldas et al. (1998) asked six literate and six illiterate women to perform word and nonword repetition tasks whilst undergoing a PET scan. In addition to finding an effect of literacy on both tasks, there were differences in the areas activated in the brain. For the literate women, there was more activation unique to the nonword repetition task, which the authors suggest represents a neural network that is involved in the processing, organisation and production of new phonological output, i.e. a nonword. The implication is that learning to read promotes explicit phonological processing and the use of this network. The ability to decode words, i.e. break words down into their constituents, is transferred to oral language and whole words are automatically split into their individual phonemes (Frith, 1998; Nation & Hulme, 2011).

The possible effect of reading accuracy on phonological skills in typically developing children has also been examined. Nation and Hulme (2011) examined the longitudinal relationship between reading accuracy and nonword repetition when children were aged 5-6 years and 6-7 years. Concurrently, reading accuracy predicted nonword repetition at the second time point, but not the first. Longitudinally, reading accuracy predicted nonword repetition and this was independent of a measure of oral language and the autoregressor. It was argued that learning to read helps create segmental phonological representations and therefore leads to more accurate repetition of nonwords.

Children with specific language impairment (SLI) typically have difficulties with phonological short-term memory and difficulties on nonword repetition tasks are seen as a cognitive marker of SLI (Hulme & Snowling, 2009). Children with SLI often also present with literacy difficulties, therefore Conti-Ramsden and Durkin (2007) examined the longitudinal relationship between these two skills in adolescents with SLI between the ages of 11 and 14 years. There was a reciprocal relationship between reading accuracy and nonword repetition, although this was not significant when the autoregressors were entered. It should be noted that there was little growth in nonword repetition over the course of the study hence it would be difficult for any variable to contribute significantly to nonword repetition after initial scores were controlled for.

1.3.3.1. Summary: Impact of reading accuracy development on oral language

There is little research on the effect that reading accuracy may have on oral language development. The studies that have been conducted focus on phonological skills and fall into two categories: those looking at the effect of life-long illiteracy and those looking at reading accuracy and oral language development in childhood. Individuals who have not received reading instruction are disadvantaged on tasks which involve segmenting spoken words or nonwords, and their patterns of brain activity suggest that, unlike adults who have learnt to read, a co-ordinated neural circuit is not activated during these tasks. During childhood, reading accuracy predicts development on nonword repetition tasks that, again, benefit from segmenting spoken stimuli. Together with the evidence reviewed above, this suggests the presence of highly interactive spoken and written language processing systems.

1.4. Levels of Cognitive Ability in Down Syndrome

Before considering the predictors of variations in reading accuracy and reading comprehension and the effect reading accuracy may have on language development in children with Down syndrome, the typical level of attainment of the predictor and outcome skills will be reviewed. The discrepancy between word and nonword reading will be described, as will the relative impairment in reading comprehension. Levels of ability in different domains of language will be summarised including phonology, grammar, vocabulary and narrative. Finally, levels of verbal and visual memory will outlined.

1.4.1. The Use of Control Groups

Control groups are typically used when conducting studies with children with Down syndrome, and indeed children with other developmental disabilities. Hodapp and Dykens (2001) outline different control groups which are frequently used in such studies: typically developing children matched on chronological or mental age, children with mixed learning difficulties, children with a different (specific) learning difficulty who show a similar target behaviour and children without learning difficulties who also show a similar target behaviour.

For children with Down syndrome, and specifically the studies which will be reviewed below, groups of typically developing children are often matched on the skill of interest to look at possible underlying differences. An example is when a control group of typically developing children matched on reading accuracy is included so differences in phonological awareness can be examined, and if differences are found these cannot be due to differences in stage of reading development. However this often leads to differences in chronological age with the children with Down syndrome being older than the typically developing children. Chronological age is important because the cognitive and behavioural profile associated with Down syndrome emerges with age (Chapman & Hesketh, 2000). Furthermore any advantage relative to a control group may be due to more experience, practice and opportunities to acquire the skill in question. Depending on the task/skill used to form a control group, differences in mental age can also emerge. This is an index of general developmental level and may affect how well children perform on tasks for non task-specific reasons, for example understanding instructions. A critical note here is that due to the uneven pattern of strengths and weaknesses in Down syndrome, the estimate of mental age can vary depending on the task used (Chapman & Hesketh, 2000). Sometimes receptive vocabulary is used, but a nonverbal task may be preferable in Down syndrome due to their relative weaknesses in verbal ability.

Children with Down syndrome can be matched for mental age and chronological age to control groups consisting of children with mixed learning difficulties. However these children are typically difficult to find, of a heterogeneous nature and due to the uneven profile associated with Down syndrome it is challenging to match the groups on every ability of interest (Chapman & Hesketh, 2000; Hodapp & Dykens, 2001).

1.4.2. Reading Accuracy in Down Syndrome.

Reading accuracy has been suggested to be a particular strength for individuals with Down syndrome, although this domain as an “island of ability” (Buckley, 1985, p. 324) has been contested. Cardoso-Martins, Peterson, Olson and Pennington (2009) examined reading accuracy in relation to verbal ability with a group of 19 individuals with Down syndrome, aged 10-19 years. Reading accuracy scores were not found to be higher than would be expected based on verbal ability, and from this the authors argued that reading accuracy is not an island of ability in Down syndrome. However, the measure of verbal ability was a receptive vocabulary task, which is a relative strength within the verbal domain (e.g. Næss, Lyster, Hulme, & Melby-Lervåg, 2011). Generally, nonverbal ability is used as an indicator of general ability and compared to this, reading accuracy has been found to be a relative strength in Down syndrome (Boudreau, 2002).

Reading accuracy is also a strength compared to other areas of cognition, including oral language and phonological awareness (Buckley, 1995; Cardoso-Martins & Frith, 2001; Roch & Jarrold, 2008; Rondal, 1995), which are predictors of reading accuracy in typical development. Given that these essential skills for learning to read are considered to be weaknesses in Down syndrome, it is perhaps surprising that reading accuracy is a relative strength. To examine whether this result may be due to chronological age differences between groups of children with Down syndrome and typically developing control groups, Fidler, Most and Guiberson (2005) compared the reading accuracy ability of a group of individuals with Down syndrome and a group of individuals with mixed learning difficulties who were matched on chronological age and nonverbal ability. There was no difference between the two groups suggesting that reading accuracy is at the same level of nonverbal ability in Down syndrome hence it is not an island of ability, and previous contradictory findings may be due to the differences in age between the children with Down syndrome and control groups. However as outlined above there are also difficulties associated with defining control groups of children with mixed learning difficulties.

1.4.2.1. Differences between word and nonword reading.

Reading accuracy is often an advantage compared to decoding in Down syndrome, for example lags of six to eight months in terms of age-equivalent scores between the two skills have been reported (Cupples & Iacono, 2000; Fowler, Doherty, & Boynton, 1995). Comparisons have also been made with typically developing control groups, for example Boudreau (2002) found that individuals with Down syndrome performed significantly better on a test of reading accuracy than a nonverbal ability matched typically developing control group, but there were no differences on a measure of decoding, which both groups scored very poorly on. The typically developing group had a mean age of 4;05, and it is therefore unsurprising that they performed at floor on the nonword reading task. It may be that comparisons on this task would be more meaningful later in development.

Roch and Jarrold (2008) administered reading tasks consisting of regular words, irregular words and nonwords to 12 individuals with Down syndrome, who were aged 10-26 years, and 14 typically developing children, who were 6-7 years. The premise behind the use of these tasks was that irregular words do not conform to normal grapheme-phoneme correspondences and therefore recruit word recognition strategies, whereas nonwords need to be decoded using grapheme-phoneme correspondences. There were no significant differences between the groups on the regular word reading task; however, the children with Down syndrome performed significantly worse on nonword reading than irregular word reading, whereas the typically developing children performed equivalently on the two tasks. Indeed the nonword reading of the children with Down syndrome was found to be impaired when their irregular word reading was accounted for. These findings strongly suggest that individuals with Down syndrome have stronger word recognition than decoding ability.

In typical development there is a regularity effect in which children show an advantage for reading regular words over irregular words as they are able to decode unfamiliar regular words. Cardoso-Martins et al.'s (2009) study investigated this in individuals with Down syndrome and found a significant regularity effect. However this effect was less pronounced than typically developing children, which supports Roch and Jarrold's (2008) finding that decoding strategies are behind word recognition ability in Down syndrome.

Roch and Jarrold (2008) argued that individuals with Down syndrome show an advantage for a visual route to reading compared to a phonological route. This was embedded in terms of Frith's (1985) stage theory, in particular the orthographic and alphabetic stages. However, this is problematic as stage theories typically require that individuals move through the stages sequentially. Therefore to have highly developed visual strategies reflecting the orthographic stage, individuals with Down syndrome should have progressed through the alphabetic stage and therefore have sufficient alphabetic strategies to read nonwords relatively well. Another way to envisage this distinction between the two reading strategies is in terms of the non-lexical and lexical routes in the DRC model, or by the phonology-orthography and phonology-semantics-orthography pathways in the triangle model, with more loading on the latter pathways in each.

An advantage for word compared to nonword reading in Down syndrome may be misleading, as it does not necessarily follow that the two skills are completely separable. Fowler et al. (1995) assessed the cognitive skills of a group of 33 individuals with Down syndrome aged 17-33 years. The group was divided into novice, emerging, developing and skilled readers on the basis of their decoding skill, and this classification predicted performance on a word reading task. This suggests that although reading accuracy is a strength in Down syndrome, it is related to nonword reading and in further support of this, significant correlations are often found between word and nonword reading (e.g. Boudreau, 2002; Cardoso-Martins et al., 2009; Cupples & Iacono, 2000; Kay-Raining Bird, Cleave, & McConnell, 2000). It must be noted that reading accuracy tasks can be completed using a variety of strategies, including both word recognition and decoding, depending upon the words within the task. Nonword reading tasks are more simplistic, having a clear load on decoding skills but knowledge of common orthographic units may also play a role. Therefore strategies in these two tasks may overlap, resulting in a significant correlation.

1.4.2.2. Components of early literacy.

Print concepts, recognition of environmental print, letter-name knowledge and letter-sound knowledge can be thought of as early or pre-literacy skills. Print concepts can be assessed by asking a child about reading conventions, for example where do you begin reading on a page and what direction do you read in? Environmental print is measured by showing children common logos or signs such as Tesco or McDonalds. Letter-sound and letter-name knowledge are often assessed simultaneously by asking children to identify letters. Of these early literacy measures, letter-sound knowledge is particularly important for the purposes of this thesis due to its role in acquiring the alphabetic principle.

Boudreau (2002) administered measures of print concepts, letter-name knowledge and letter-sound knowledge to individuals with Down syndrome and typically developing children of the same nonverbal ability. Despite having better reading accuracy skills, the individuals with Down syndrome performed equivalently on all three tasks of early literacy compared to the typically developing group. However it is perhaps more informative to compare groups at the same reading accuracy level to investigate whether these skills are comparable. In their first experiment Snowling , Hulme and Mercer (2002) found no such difference on tasks of letter-name knowledge, letter-sound knowledge and environmental print between a group of children with Down syndrome and a reading accuracy-matched typically developing group. In contrast, in a subsequent experiment letter-name knowledge was found to be at the same level in the two groups whereas the children with Down syndrome performed more poorly than the typically developing children on the letter-sound knowledge task. The authors accounted for the conflicting findings by suggesting that the two groups of children with Down syndrome who participated in the respective studies may have received different literacy instruction. Cardoso-Martins and Frith (2001) also found conflicting results with no differences between children with Down syndrome and a reading accuracy-matched typically developing group on capital letter-name knowledge, but there was an advantage for the typically developing group on a lower-case letter-name knowledge task.

1.4.2.3. Summary: Reading accuracy in Down syndrome.

Down syndrome is characterised by an uneven cognitive profile in which reading accuracy is a relative strength. Early literacy skills appear to be relatively well-developed but there are mixed results as to whether letter knowledge is in line with reading accuracy. When considering sub-skills of reading in more detail, word and nonword reading are related in Down syndrome, but there appears to be a disparity between the two with higher levels of reading accuracy than decoding ability. This is likely to be due to poor phonological awareness skills, which will be discussed in more detail below. It is also relevant to note that older individuals with Down syndrome such as those studied by Roch and Jarrold (2008) or those involved in less recent studies are likely to have been taught to read using primarily visual strategies, which would presumably result in an advantage for familiar words over words that cannot be read visually, i.e. nonwords.

1.4.3. Reading Comprehension in Down Syndrome

Reading comprehension has been highlighted as an area of difficulty within the literacy domain for individuals with Down syndrome, with levels of comprehension lagging behind those of reading accuracy (Cardoso-Martins et al., 2009; Fletcher & Buckley, 2002; Nash & Heath, 2011; Verucci, Menghini, & Vicari, 2006). This is particularly apparent in children with Down syndrome who are high-ability readers; Nash and Heath (2011) found that children with Down syndrome reading at an age-equivalent level of 10;02 had an average discrepancy with reading comprehension of 33 months.

Groen, Laws, Nation and Bishop (2006) reported a case study of KS, a girl with Down syndrome who had exceptional reading accuracy skills. She had a discrepancy between reading accuracy and comprehension which was larger than is seen in typical development but comparable to that seen in poor comprehenders. Similarly Nash and Heath (2011) found that, with only one exception, their sample of 13 children with Down syndrome all had a discrepancy of at least 12 months between their reading accuracy and comprehension. The children with Down syndrome were compared to a group of poor comprehenders, and they had similar profiles on tasks of reading comprehension, reading accuracy and receptive vocabulary, although the children with Down syndrome scored more poorly on decoding and inferential questions.

1.4.4. Oral Language Skills in Down Syndrome

It is well-documented that children with Down syndrome have delayed oral language development compared to their nonverbal abilities (e.g. Fowler, 1990; Fowler 1995; Rondal, 1995). Children with Down syndrome present with difficulties in various language domains including phonology, syntax, morphology, semantics and pragmatics (Rondal, 1995), however it is apparent that the degree of deficit in each domain varies greatly.

There is a distinction between expressive language, i.e. the ability to produce language, and receptive language, i.e. the ability to understand language. Typically, children with Down syndrome have deficits in expressive language compared to both their nonverbal ability and receptive language; a profile which is evident from a young age and continues throughout development (Cleland, Wood, Hardcastle, Wishart, & Timmins 2010; Jenkins, 1993; Laws & Bishop, 2003; Miller, Leddy, Miolo, & Sedey 1995).

This section will outline typical levels of attainment in the domains of phonology, grammar, vocabulary and narrative, and relative strengths and weaknesses within the language domain. The distinction between expressive and receptive language will also be discussed.

1.4.4.1. Phonology.

Articulation or speech production tasks can be used to assess how well children can produce phonemes in different positions in a word. These often take the form of picture naming tasks, from which several measures can be calculated including percentages of consonants, vowels and total phonemes produced correctly. Errors on tasks such as this can be typical, i.e. they occur in typical development, or atypical, i.e. they do not normally occur in typical development. The former would indicate delayed phonology, whereas the latter would indicate disordered phonology. Intelligibility can also be calculated from this elicited production of target words or from a language sample.

Phonological awareness tasks are used to measure how well children can explicitly reflect on sounds within words. In individuals with Down syndrome, there has been comparison between rime and phoneme awareness and therefore tasks which focus on different units of phonological awareness are commonly used. Tasks of different complexity, for example recognition vs. deletion, are also used.

Memory for phonological representations is often assessed by asking children to repeat nonwords. If children are better able to temporarily store new phonological forms then they will be more successful on this task; however poor hearing, speech discrimination or articulation would also impact on performance.

1.4.4.1.1. Speech production and discrimination.

Smith and Stoel-Gammon (1983) investigated early phonological development in children with Down syndrome by studying the production of stop consonants (p, t, k, b, d, g) longitudinally. Between the age of three and six years children with Down syndrome made less progress than younger typically developing children. To give some indication of the delay, when children with Down syndrome were aged three years they performed at the level of a 1-2 year old typically developing child, and this discrepancy increased with age. However the two groups were not matched on any variable, therefore it is difficult to judge the degree of the phonological delay.

Roberts et al. (2005) investigated the articulation skills of children with Down syndrome relative to their nonverbal ability by comparing the articulation of individual words in 32 children with Down syndrome aged 4-13 years, and a group of 33 younger typically developing children aged 2-6 years matched on nonverbal ability. Children with Down syndrome were less accurate at producing individual consonants and therefore their attempts at whole words resembled the target words less well than the typically developing children. Similar results in connected speech were found by Barnes et al. (2009); however many participants had also taken part in Roberts et al., so similar results are perhaps unsurprising.

It appears that the articulation skills of children with Down syndrome are a weakness compared to nonverbal ability. Although children with Down syndrome made more errors than typically developing children in the studies of Roberts et al. (2005) and Barnes et al. (2009), the types of errors they made were similar. Furthermore Kumin, Councill and Goodman (1994) described the order of acquisition of phonemes in the speech samples of 60 children with Down syndrome aged nine months to nine years, and the reported sequence is similar to that reported in a normative study of typically developing children (Dodd, Holm, Hua, & Crosbie 2003). This suggests that phonological development in Down syndrome is delayed, but not disordered. However in contrast to this, Cleland et al. (2010) found that two-thirds of articulation errors produced by their sample of 15 children with Down syndrome were classified as delayed, but all children made at least one error which represented disordered phonology. This suggests that some elements of speech may be disordered in Down syndrome.

To clarify whether speech difficulties associated with Down syndrome should be classified as delayed or disordered, Dodd and Thompson (2001) compared individuals with Down syndrome aged 5 -15 years to otherwise typically developing children aged 3-5 years who had a phonological disorder characterised by inconsistencies. There were 15 individuals in each group pairwise matched on the percentage of correct phonemes produced in an articulation test. Phonological inconsistency was measured by administering a picture naming task three times and comparing responses on each occasion. It was found that a similar number of words were produced inconsistently in the two groups, which suggests that the phonological development of children with Down syndrome is disordered and is characterised by the inconsistent realization of phonemes.

As children with Down syndrome have difficulty producing correct phonemes, it is likely that this affects how well their speech can be understood. Barnes et al. (2009) found that children with Down syndrome produced less intelligible speech than typically developing children of the same nonverbal ability, although levels of intelligibility were relatively high, i.e. above 80%, for both groups. In comparison, Cleland et al. (2010) found that the average amount of intelligible speech produced by children with Down syndrome was 52%, although there was a large degree of variability. These children were all taking part in a larger intervention study which aimed to improve intelligibility, and parents of children with poor intelligibility would be more likely to volunteer for such a study, which may explain the discrepancy with Barnes et al.

Keller-Bell and Fox (2007) examined the speech discrimination skills of eight individuals with Down syndrome aged 5-12 years, matched to a group of seven typically developing children aged 4-5 years on nonverbal ability. Speech discrimination was tested using five synthetic stimulus pairs (e.g. ba-da). The children with Down syndrome performed worse than the typically developing children on two out of the five pairs, which suggests that individuals with Down syndrome have problems discriminating some speech contrasts. Furthermore these results are unlikely to be explained by hearing status as only children who had hearing losses less than 25db were included. If individuals with Down syndrome are experiencing problems discriminating speech sounds, it is perhaps unsurprising that they have difficulties in phoneme production.

1.4.4.1.2. Phonological awareness.

One of the first studies to look at phonological awareness in individuals with Down syndrome was Cossu, Rossini and Marshall (1993). They administered tasks of phoneme segmentation, phoneme blending and onset deletion to 10 children with Down syndrome aged 8-15 years and 10 typically developing children aged 6-7 years who were matched on reading accuracy. The children with Down syndrome performed significantly worse than the typically developing children on all the phonological awareness tasks. From these results, Cossu et al. argued that phonological awareness is not involved in reading for individuals with Down syndrome and thus phonological awareness is not a necessary prerequisite for learning to read. However many flaws with this study have been highlighted, including a large degree of variability in the phonological awareness scores of the children with Down syndrome, which showed that some of the children had measurable levels of phonological awareness (Byrne, 1993). Furthermore Cardoso-Martins and Frith (2001) argued that some of the tasks used by Cossu et al. were very cognitively demanding. Indeed their phoneme blending task included items with six phonemes; as individuals with Down syndrome have low verbal memory spans, typically lower than six (Kay-Raining Bird & Chapman, 1994), it is perhaps unsurprising they would find this task difficult. Thus the children with Down syndrome may have found these tasks difficult for reasons other than having poor phonological awareness.

It seems that this early research into the phonological awareness skills of children with Down syndrome reached prematurely negative conclusions due to methodological flaws. More recent research has illustrated that individuals with Down syndrome can indeed demonstrate measurable levels of phonological awareness, and this will now be discussed.

Kennedy and Flynn (2003) investigated the levels of phonological awareness in a group of nine children with Down syndrome aged 5-8 years. Rime detection, alliteration detection, initial phoneme isolation and phoneme blending were assessed, using tasks which required a multiple-choice nonverbal response. There was a range of abilities within the group; some children did not score above chance on any task whilst some children scored above chance on multiple tasks. Although this study only included nine children, it showed that individuals with Down syndrome can perform above chance on phonological awareness tasks.

Many studies focussing on phonological awareness and reading accuracy have included typically developing control groups and larger sample sizes. Cardoso-Martins and Frith (2001) matched 33 individuals with Down syndrome and 33 typically developing children on reading accuracy and investigated their phonological skills. The typically developing group were aged 6-9 years, whereas the individuals with Down syndrome ranged from 10-49 years. The typically developing children performed significantly better on an initial phoneme deletion task but both groups performed close to ceiling on an easier task of initial phoneme detection. It appears, therefore, that some elements of phonological awareness are less developed in individuals with Down syndrome in comparison to reading accuracy while others may be in line. However due to the large age range of the individuals with Down syndrome there is likely to be great variation in the type of schooling, and therefore reading instruction, experienced.

Other studies have also utilised reading accuracy matched control groups with different phonological tasks, and the results appear to converge on Cardoso-Martins and Frith's (2001) findings. Snowling et al. (2002) found that individuals with Down syndrome performed significantly poorer on measures of syllable segmentation and phoneme detection than a reading accuracy matched typically developing control group. However, these differences were no longer significant when receptive vocabulary, used as an index for general ability, was controlled for. In a second experiment, a phoneme detection task was administered which targeted both initial and final phonemes. The two groups performed similarly on the initial phoneme detection task but there was a trend for the children with

Down syndrome to score more poorly than the typically developing children on the final phoneme detection task. However in this second experiment, the group of children with Down syndrome were better readers than the typically developing children. In Roch and Jarrold's study (2008) there was no difference on a phoneme detection task between individuals with Down syndrome and a reading accuracy matched group of typically developing children, although the typically developing group did perform significantly better on measures of phoneme deletion. Therefore it appears that individuals with Down syndrome do have difficulties with phonological awareness but this is not consistent across studies.

The inconsistency of findings may be due to the different phonological awareness measures used by different studies, as these tasks can vary in their degree of complexity. Individuals with Down syndrome appear to be relatively unimpaired compared to their reading accuracy skill on simple tasks such as phoneme matching (Fletcher & Buckley, 2002, Cardoso-Martins & Frith, 2001; Roch & Jarrold, 2008; Snowling et al., 2002). Performance on tasks requiring a greater degree of manipulation, such as blending and segmentation, is less clear-cut, with conflicting results (Boudreau, 2002; Cossu et al., 1993; Fletcher & Buckley, 2002). However, difficulties with the more complex task of phoneme deletion have been consistently found (Cardoso-Martins & Frith, 2001; Cossu et al., 1993; Roch & Jarrold, 2008). Therefore it appears that individuals with Down syndrome may have lower-level phonological skills commensurate with their reading accuracy, but tasks requiring a greater degree of manipulation prove more challenging.

Phonological awareness tasks can assess different units of sound: whole words, syllables, onsets and rimes or phonemes. Snowling et al. (2002) administered alliteration and rime tasks, designed to have the same format and to have relatively low cognitive demands. The group of children with Down syndrome only scored above chance in the alliteration task, whereas the reading accuracy matched typically developing control group scored above chance in both tasks. This particular deficit in rime has been replicated in other studies (e.g. Cardoso-Martins, Michalick, & Pollo 2002; Gombert, 2002) and raises the possibility that children with Down syndrome may be biased towards the initial sounds of words, especially as relative weaknesses have also been found on tasks focussing on final phonemes (Snowling et al., 2002).

Children with Down syndrome typically have lower mental ages than reading accuracy matched typically developing control groups, which may result in more difficulty understanding task demands. This can be tested by comparing the phonological awareness skills of individuals with Down syndrome and typically developing children matched on nonverbal ability. Boudreau (2002) administered a large test battery to 20 children with Down syndrome aged 5-17 years and 20 typically developing children aged 3-5 years matched on nonverbal ability. It was found that individuals with Down syndrome performed at a similar level as nonverbal ability matched typically developing children on measures of blending and syllable segmentation, but the typically developing group performed significantly better on tasks of rime judgement and alliteration judgement. This study suggests that the difficulties with phonological awareness typically seen in children with Down syndrome are not wholly due to their lower mental age.

1.4.4.1.3. Phonological memory.

Nonword repetition is a complex task and involves many processes that are typically a weakness for children with Down syndrome, including hearing, speech discrimination, planning speech-motor output and articulation. Therefore the first studies administering this task to individuals with Down syndrome aimed to establish whether nonword repetition could be used reliably. Laws (1998) assessed the repetition of words and nonwords of different lengths with 33 children with Down syndrome aged 5-18 years and found that they were less accurate on longer items than shorter items, which mirrors the well-established length effect found in typical development (e.g. Archibald & Gathercole, 2007; Gathercole et al., 1991). Comblain (1999) also found evidence of the length effect in a group of individuals with Down syndrome, along with the wordlikeness effect which is also present in typical development (Gathercole et al., 1991). This suggests that nonword repetition is tapping the same underlying processes in children with Down syndrome as in typical development.

The performance of children with Down syndrome compared to typically developing children has been investigated by Cairns and Jarrold (2005) and Keller-Bell and Fox (2007). Cairns and Jarrold compared a group of 18 individuals with Down syndrome aged 12-19 years and typically developing children aged 5-7 years who had similar levels of vocabulary. They found that the individuals with Down syndrome performed significantly worse on a measure of nonword repetition than the typically developing children, but there was an equivalent wordlikeness effect in both groups. These findings were replicated by Keller-Bell and Fox with a control group of children matched on nonverbal ability. It should be noted that in Cairns and Jarrold, the typically developing children performed significantly better than the children with Down syndrome on an articulation task, which may have affected their performance. In summary, the nonword repetition skills of children with DS appear to be a difficulty beyond their vocabulary and nonverbal skills.

1.4.4.2. Grammatical skills.

Grammar can be subdivided into syntax and morphology. Syntax refers to the rules which guide the combination of words into phrases and sentences. Receptive syntax is often measured by assessing whether a child can correctly identify pictures that represent sentences, which increase in complexity. The complexity of a child's spoken language is often measured indirectly with their mean length of utterance (MLU; Dethorne, Johnson, & Loeb, 2005). This is often calculated from a language sample or a narrative, or parents can be asked to estimate their child's MLU, especially in early development. MLU correlates highly with other measures of grammatical complexity at a young age (Rice, Redmond, & Hoffman, 2006) but it is argued to become less representative of expressive syntax later in development (Scarborough, Rescorla, Tager-Flusberg, Fowler, & Sudhalter 1991). Morphemes are the smallest units of meaning, which can include individual words (free morphemes) but also include affixes, suffixes, inflections and derivations. Morphology can be assessed with receptive tasks but is more commonly assessed expressively in groups of individuals with Down syndrome by analysing speech samples or using tests to elicit target morphemes. Studies focussing on the syntactic and morphological skills of individuals with Down syndrome will now be discussed.

Receptive syntax can easily be assessed in children with Down syndrome as no production response is required. Joffe and Varlokosta (2007) used receptive tasks of general syntax and more specific constructs such as active and passive sentences and wh-questions (who, what and which) with 10 children with Down syndrome aged 5-14 years and 10 typically developing children aged 3-6 years, who were matched for nonverbal ability. Children with Down syndrome were impaired relative to the typically developing children on all tasks, with the exception of the simpler active sentences. Vicari, Carlesimo and Caltagirone (2000) used a different task, which assessed children's ability to act out spoken instructions that increased in complexity. Fifteen children with Down syndrome aged 4-7 years performed more poorly on this task than typically developing children matched on nonverbal ability. Therefore individuals with Down syndrome appear to present with receptive syntax difficulties relative to nonverbal ability, and this is found on different tasks and at different stages in development (see also Laws & Bishop, 2003; Price, Roberts, Vandergrift, & Martin 2007), and this pattern of results was also confirmed by a recent meta-analysis by Næss et al. (2011).

Due to the general weaknesses in expressive language in children with Down syndrome along with their relatively poor receptive syntax, difficulties in producing complex sentences would also be expected. Singer-Harris, Bellugi, Bates, Jones and Rossen (1997) used the MacArthur Communicative Development Inventory to assess the language abilities of a group of children with Down syndrome and a group of children with Williams syndrome, with mean ages of 3;11 and 2;09 respectively. This inventory is a parental questionnaire, which includes questions regarding the length and complexity of a child's speech and it was found that the children with Down syndrome scored more poorly than the children with Williams syndrome.

The children with Williams syndrome were not matched on any variable to the children with Down syndrome in Singer-Harris et al.'s (1997) study, so they could be at a different developmental stage. Vicari et al. (2000) more directly evaluated the early development of syntax by including a control group of typically developing children matched on nonverbal ability. The Italian version of the MacArthur Communicative Development Inventory was used to collect information about children's speech and MLU was calculated from the child's spontaneous production during the assessment. More children with Down syndrome were reported by parents to use simpler sentences and sentences omitting function words than typically developing children. Furthermore the children with Down syndrome had lower MLUs, supporting the parental reports. Deficits in expressive syntax have also been found in older individuals (e.g., Boudreau & Chapman 2000; Buckley, 1995; Chapman, Seung, Schwartz, & Kay-Raining Bird 1998; Fabbretti, Pizzuto, Vicari, & Volterra, 1997; Price et al., 2008).

MLU is a broad and indirect measure of sentence complexity and to investigate syntax use more explicitly, sentences or clauses can be analysed in depth for their complexity. Thordardottir, Chapman and Wagner (2002) categorised the syntax produced in sentences by individuals with Down syndrome aged 12-20 years and typically developing children aged 2-4 years who had a similar MLU. The complexity of the syntax produced was equivalent in the two groups, suggesting that MLU does reflect complex syntax use. Furthermore different categories of complex sentences emerged in a similar sequence in the two groups, when the groups were split cross-sectionally by MLU level. In contrast Fabbretti et al. (1997) found that children with Down syndrome aged 6-15 years produced more simple clauses than typically developing children aged 2-6 years matched for MLU. These two studies were carried out in different languages and with different age groups of children, which perhaps may account for the differences in results.

Scarborough et al. (1991) investigated how well MLU can be used to predict syntactic complexity in children with Down syndrome. The Index of Productive Syntax (IPSyn) measures grammatical complexity of speech by scoring language samples for the presence of 56 syntactic and morphological forms. In a cross-sectional sample of 30 typically developing children aged 2-4 years and a longitudinal sample of five children with Down syndrome aged 4-5 years, the correlation between MLU and IPSyn was very strong. However for both groups of children, as MLU increased, the correlation weakened and MLU over-estimated IPSyn especially for the children with Down syndrome. This suggests that individuals with Down syndrome produce long but syntactically simple sentences, especially in later development.

There are different components within a sentence which affect syntax, including function words, verbs and nouns along with the order of words. Verb production has been specifically examined and it appears that individuals with Down syndrome have difficulties in this domain beyond their MLU. Hesketh and Chapman (1998) found that compared to typically developing younger children matched on MLU, adolescents with Down syndrome produced fewer verbs per utterance. This difference appeared to be due to an increased number of utterances without a verb in the group of individuals with Down syndrome. Similarly Grela (2003) found more errors in verb phrases in a group of seven children with Down syndrome aged six to twelve years than typically developing children matched on MLU.

Early morphological skills have been studied, particularly focussing on the order of acquisition of grammatical markers. In a longitudinal study, Rutter and Buckley (1994) found great variability in parental reports of the acquisition order of morphemes of 12 children with Down syndrome aged 1-3 years. No typically developing control group were included but the results from the children with Down syndrome were compared to norms for typically developing children described in Brown (1973). The children with Down syndrome acquired some morphemes in a different sequence to typically developing children, and others were omitted completely, suggesting that there may be differences in morphological development from an early age.

The large degree of variability in Rutter and Buckley's (1994) study and the small sample size makes generalisation difficult. Berglund, Eriksson and Johansson (2001) collected parental reports on oral language in a cross-sectional nationwide study of Swedish children with Down syndrome. Three hundred and thirty children with Down syndrome aged 1-5 years participated and their performance was compared to existing data from 336 typically developing children aged 1-2 years. It was found that the acquisition of morphemes occurred in a similar order in both groups of children. Berglund et al. also examined the frequency of morpheme use. When matched for vocabulary size, typically developing children were reported to use morphemes, specifically possessive nouns/pronouns, singular definite forms and singular indefinite forms, more often than children with Down syndrome. Similarly, for older children with Down syndrome aged 8-18 years, Bol and Kuiken (1990) found that morphemes indicating verb agreement and the past tense occurred more frequently in typically developing children aged two to three years.

Within grammar, there is evidence to suggest that children with Down syndrome have difficulties with morphology relative to syntax. Eadie, Fey, Douglas and Parsons (2002) compared the speech samples of 10 children with Down syndrome, who had a mean age of 7;03 and 10 typically developing children who had a mean age of 3;04 matched for MLU. Compared to the typically developing group, the children with Down syndrome omitted more morphemes in spontaneous speech. This has been replicated relative to MLU (Fabbretti et al., 1997; Fowler, Gelman, & Gleitman 1994) and extended to comparisons with control groups matched on receptive syntax (Boudreau & Chapman, 2000; Miles, Chapman, & Sindberg, 2006). In support of this distinction, Abbeduto et al. (2003) found that the correlation between receptive tasks of morphology and syntax was close to zero for individuals with Down syndrome but moderate to strong for typically developing children.

1.4.4.3. Vocabulary.

Expressive vocabulary can be measured using picture naming tests or by calculating the total number of words or the number of different words produced in speech, the latter being a measure of lexical diversity. Expressive vocabulary can also be assessing using definition tasks, but this has not been used widely with individuals with Down syndrome presumably because of the potential impact of expressive language difficulties. Receptive vocabulary is often measured with multiple-choice tasks that require children to choose the picture which matches a word.

The early development and onset of vocabulary was investigated as part of Berglund et al.'s (2001) large cross-sectional study of children with Down syndrome aged one to five years, and most children uttered their first word by the age of two years, although there was a large amount of variability. Some of the children with Down syndrome began to talk at the same age as typically developing children, suggesting that the onset of spoken word production is not necessarily delayed. However the rate of acquisition was slower, with four year olds with Down syndrome performing equivalently to typically developing children aged eighteen months. Stefanini, Caselli and Volterra (2007) explored the expressive vocabulary ability in early childhood using a picture naming task. Fifteen children with Down syndrome aged 3-8 years took part, along with an age-matched typically developing group and a mental age-matched typically developing group aged 2-4 years. The children with Down syndrome had poorer expressive vocabulary knowledge than either control group, which suggests that after the onset of speech, early vocabulary development is delayed supporting Berglund et al.

In a large group of individuals with Down syndrome aged 5-20 years, Chapman et al. (1998) found that the total number of words and number of different words produced in language samples were lower compared to typically developing children at the same level of nonverbal ability. This finding has been replicated with the same group of individuals six years later (Boudreau & Chapman, 2000) and also with younger children with Down syndrome

(Miller, 1988). Fidler et al. (2005) used a receptive vocabulary task with a group of children and adolescents with Down syndrome and a group of individuals with learning difficulties matched on nonverbal ability and age. Again, a relative weakness in vocabulary was found for the individuals with Down syndrome.

The studies reviewed above suggest vocabulary is impaired relative to nonverbal ability; however other studies have found that these two skills are in line with each other. Laws and Bishop (2003) compared the language skills of 19 participants with Down syndrome aged 10-19 years and typically developing children aged 4-7 years of the same nonverbal ability. Although there was a trend for the individuals with Down syndrome to perform more poorly than the typically developing children on receptive vocabulary and picture naming tasks, this was not significant. Indeed their receptive vocabulary standard score calculated using mental ages rather than chronological age was well within the normal range at 96. Other studies have also found that receptive vocabulary is in line with nonverbal ability (e.g. Chapman, Kay-Raining Bird, & Schwartz, 1990; Cleland et al., 2010; Jarrold, Thorn, & Stephens, 2009; Mosse & Jarrold, 2011). Laws and Bishop's findings for expressive vocabulary have also been replicated with picture naming tasks (Chapman et al., 1990; Jarrold et al., 2009) and lexical diversity (Vicari et al., 2000).

The findings are therefore conflicting regarding whether vocabulary is in line or below nonverbal ability. Næss et al.'s (2011) meta-analysis highlights the importance of considering receptive and expressive skills separately as it was found that across a number of different studies, children with Down syndrome performed significantly poorer than nonverbal ability matched control groups on expressive vocabulary tasks, but not receptive vocabulary.

1.4.4.4. Grammar and vocabulary asynchrony.

Grammar is consistently impaired compared to nonverbal ability in children with Down syndrome, whereas the evidence for vocabulary is mixed, largely dependent on whether the task is expressive or receptive. The implication from this is that grammar may be more impaired than vocabulary.

This asynchrony between the development of grammar and vocabulary appears to be present from an early age. Singer-Harris et al. (1997) found that children with Down syndrome with a mean age of 3;03 showed a greater dissociation between parental reports of lexical and grammatical development than would be expected based on normative data. Similarly, Vicari et al. (2000) found that compared to a typically developing control group matched on mental age children with Down syndrome aged 4-7 years did not differ on a parental measure of lexical diversity but produced less grammatically complex speech.

This dissociation is also evident in later childhood, with a variety of different vocabulary and grammar measures. Miller (1988) found that in comparison to typically developing children with equivalent MLU, children with Down syndrome aged 2-12 years had greater lexical diversity in a language sample. Similarly Laws and Bishop (2003) found that individuals with Down syndrome aged 10-19 years did not differ from a typically developing control group matched on nonverbal ability on measures of receptive and expressive vocabulary but did perform significantly poorer on measures of both expressive and receptive syntax. Furthermore within a group of children and young adults with Down syndrome, Chapman, Schwartz and Kay-Raining Bird (1991) found an advantage for receptive vocabulary compared to receptive syntax, which was equivalent to a one year discrepancy in age-equivalent scores and was not present for typically developing children.

1.4.4.5. Narrative ability.

Narratives offer rich sources of data, apart from the most common measures of MLU or lexical diversity. The child's ability to understand and/or express the key themes, or events, of the narrative can be measured by asking children to narrate sequences of pictures, retell previously heard narratives or answer questions about the story. Narratives can be presented orally and/or visually and to perform well the child needs to attend to this, identify the key points, synthesise them and construct an overall representation. Therefore this is a higher-order multi-componential skill. For children with Down syndrome, an additional complication may be remembering this information. Narrative comprehension or production is therefore a linguistically and conceptually complex task, which may be limited by memory and expressive language demands in Down syndrome.

Loveland, McEvoy and Tunali (1990) compared 16 individuals with Down syndrome and 16 individuals with autism aged 5-27 years on a narrative retelling task. The two groups were matched on general verbal ability, but the individuals with Down syndrome performed significantly poorer on a measure of nonverbal ability. The two groups saw a visually presented story and on a free recall task they recalled a similar number of events. This suggests that the narrative skills of children with Down syndrome are in line with their general language ability, but may be below their nonverbal ability. However, the individuals with autism performed significantly worse on the pragmatic aspects of narrative re-telling, for example expressing the narrative as a cohesive whole, and question answering, and may not be the most representative control group. Furthermore children saw different stories, either played out by puppets or on video, depending on their age.

Boudreau and Chapman (2000) included three different control groups matched on nonverbal ability, syntax comprehension and MLU to investigate which of these skills narrative ability was commensurate to in children and adolescents with Down syndrome. Children's narrative descriptions of a silent video were analysed for the number of different events included. The individuals with Down syndrome performed equivalently to the typically developing groups matched on nonverbal ability and syntax comprehension, but significantly better than the MLU-matched group. Similar results were found by Miles and Chapman (2002) who analysed narratives produced in response to a picture book using the same matching design and some of the same sample. Together, these results suggest that individuals with Down syndrome can express their understanding of a sequence of events in line with their general ability level. The results regarding the different control groups suggest that although individuals with Down syndrome are able to express narrative content to the same level as their syntactic comprehension, they may produce simpler language to do so (Miles & Chapman, 2002).

1.4.4.6. Impact of hearing and speech-motor skills on oral language

Permanent and temporary hearing losses are common in people with Down syndrome and it has been argued that this may affect future language proficiency (Abbeduto et al., 2007). In a series of studies with the same group of children, Chapman and colleagues (Chapman et al., 1991; Chapman, Seung, Schwartz, & Kay-Raining Bird 2000) found that hearing predicted unique variance in receptive and expressive language even after controlling for nonverbal ability and age, although this was low at approximately 3-8%. Within grammar, Miolo, Chapman and Sindberg (2005) found that hearing accounted for 23% of variance in a receptive morphology task, but no unique variance in a receptive syntax task. Furthermore Laws and Bishop (2003) found that within morphology, hearing loss was not related to the use of third person singular morphemes but there was a significant relationship with past tense morphemes.

In contrast, hearing has also been found not to correlate with receptive and expressive language; Laws (2004) found that hearing did not correlate significantly with MLU or lexical diversity and Marcell and Cohen (1992) found that when the effects of age and IQ were controlled for, hearing level did not significantly correlate with receptive and expressive language skills. In summary, there are very mixed findings on whether hearing level is related to language skills in Down syndrome, which does not seem to be due to the use of different tasks.

There is comparatively little research on the relationship between speech-motor skills and language in Down syndrome. A recent study by Cleland et al. (2010) examined the correlation between speech-motor skills, intelligibility and articulation with 15 individuals with Down syndrome aged 9-18 years. All three domains were highly intercorrelated, suggesting that those children with poor speech-motor skills had greater impairments in intelligibility and articulation. However these measures did not correlate with other language measures, suggesting that difficulties in speech and broader language are relatively independent.

1.4.4.7. Summary: Oral language skills in Down syndrome.

There has much research into the oral language skills of children with Down syndrome. Before summarising the findings, it is necessary to highlight the fact that the data from one sample of children has been used in many studies by the same research group (e.g. Hesketh & Chapman, 1998; Miolo et al., 2005; Chapman et al., 1991; Chapman et al., 2000), and therefore it is likely that the results from these studies would be similar.

Children with Down syndrome present with difficulties on phonological tasks compared to nonverbal ability. This includes difficulties with speech discrimination, articulation and nonword repetition tasks. If children have difficulty discriminating phonemes, then this may result in the formation of imprecise phonological representations and therefore inaccurate and inconsistent articulation. This also has the potential to impact on phonological processing skills and therefore literacy development. Despite early research arguing that individuals with Down syndrome do not possess phonological awareness, the weight of evidence suggests that individuals with Down syndrome do find phonological manipulation difficult but are able to perform above chance on some tests, although rime awareness and complex tasks such as deletion appear to be particularly challenging.

The domain of grammar is also a particular weakness for individuals with Down syndrome relative to nonverbal ability. Within grammatical skills, as reflected in oral language overall, children with Down syndrome perform more poorly on expressive tasks compared to receptive. There is also some evidence that the correct use of morphemes is impaired beyond both expressive and receptive measures of syntax.

The age of onset of spoken word production appears to be similar in children with Down syndrome as typically developing children, although there is much variability. The rate of development however, is slower and this difference begins in the early years and continues throughout childhood. When compared to children of the same nonverbal ability, receptive vocabulary appears to be unimpaired, whereas expressive vocabulary is poorer. This is consistent with the discrepancy between receptive and expressive language commonly seen in Down syndrome. There is an advantage for vocabulary relative to grammar for individuals with Down syndrome throughout development. However the two domains have been found to be significantly correlated, suggesting that they are not completely dissociated (Vicari et al., 2000).

The ability of individuals with Down syndrome to express the key content of narratives is in line with their understanding of syntax and better than expected given their expressive language. Perhaps due to their poorer expressive language, they use simpler language to express the same content as typically developing children matched for receptive syntax. However, both Boudreau and Chapman (2000) and Miles and Chapman (2002) used the same sample of children with Down syndrome and therefore more independent studies are needed to confirm these findings.

1.4.5. Memory in Down Syndrome

Poor verbal relative to visual skills are generally considered part of the cognitive profile associated with Down syndrome. This distinction has been followed in studies of memory in individuals with Down syndrome. A clear example of this is a study by Jarrold et al. (1999), which compared 25 individuals with Down syndrome, 16 individuals with Williams syndrome and 17 individuals with moderate learning difficulties on tasks of verbal and visuospatial short-term memory. The groups had mean ages of 12;07, 16;09 and 12;05 respectively. On a digit recall task, the children with Down syndrome were impaired compared to both other groups of children, even when accounting for performance on Corsi blocks, nonverbal ability and receptive vocabulary, which was used as an index of general verbal ability. Conversely, the individuals with Williams syndrome performed significantly worse on the Corsi blocks task than the individuals with Down syndrome and the individuals with moderate learning difficulties when differences in digit recall along with verbal and nonverbal ability were taken into account. Therefore the individuals with Down syndrome showed an impairment in verbal short-term memory, which did not extend to visuospatial short-term memory. This finding is robust and has been replicated across different studies (e.g. Frenkel & Bourdin, 2009; Jarrold & Baddeley, 1997; Lanfranchi, Baddeley, Gathercole, & Vianello, 2009; Lanfranchi, Carretti, Spano, & Cornoldi, 2004; Laws, 2002). Furthermore problems in hearing, encoding of auditory stimuli and speech output demands do not seem to underlie this deficit in the phonological loop (Laws, 2002).

In comparison to the deficits found in verbal short-term memory, children with Down syndrome generally perform at an equivalent level as typically developing children of the same nonverbal ability on tasks of visuospatial short-term memory (Hick, Botting, & Conti-Ramsden, 2004; Lanfranchi et al., 2004; Visu-Petra, Benga, Tincas, & Miclea 2007). It has been suggested that it is the spatial component of visuospatial memory which individuals with DS are superior on (e.g. Laws, 2002). Indeed Frenkel and Bourdin (2009) found a relative deficit on a visual short-term memory task, but not spatial short-term memory for individuals with Down syndrome relative to typically developing children of a similar nonverbal ability.

The ability of children with Down syndrome to simultaneously store and process information (i.e. working memory) has also been investigated. Deficits in verbal tasks would be expected because of impairments in the temporary storage of verbal information. If impairments were also found on visuospatial tasks this would suggest a deficit with domain general processing i.e. the central executive. Lanfranchi et al. (2004) examined the effect of increasing the processing demands of verbal and visuospatial memory tasks. In both domains the difference in scores between children with Down syndrome and typically developing children of the same nonverbal ability increased as the difficulty of the task increased. This suggests that children with Down syndrome have difficulties with domain-general processing demands. In a later study Lanfranchi et al. (2009b) confirmed this pattern of results with children with Down syndrome and typically developing children matched on verbal ability and vocabulary. Therefore the working memory deficit does not appear to be due to impaired general ability or language skills.

1.5. Predictors of Reading Accuracy, Reading Comprehension and Language Development in Down Syndrome

1.5.1. Predictors of Reading Accuracy in Down Syndrome

1.5.1.1. Relationship between reading accuracy and memory.

It has been suggested that relative strengths in the visual domain may partially explain strengths in word recognition among children with Down syndrome, as they can recognise the shape of a familiar word, which then activates the associated pronunciation (Boudreau, 2002; Buckley, 1985; Kay-Raining Bird, Cleave, & McConnell, 2000). There have been many studies exploring the reading accuracy of individuals with Down syndrome, but few of these include measures of visual processing and/or memory. One exception is a study by Fidler et al. (2005), who compared the relationship between visual processing and reading accuracy in a group of adolescents with Down syndrome compared to a chronological age and nonverbal ability matched group of adolescents with mixed learning difficulties. The groups also performed at an equivalent level on measures of reading accuracy and visual processing. After accounting for chronological age, visual processing was found to contribute a significant proportion of variance to reading accuracy in the individuals with Down syndrome but not the individuals with mixed learning difficulties, which supports the proposal that visual skills aid reading accuracy for individuals with Down syndrome. Fidler et al. suggested that a sight word approach to reading should be encouraged based on these findings. However this conclusion could be considered premature from a practical point of view, as this minimises the importance of being able to decode a new word, and also from a theoretical point of view, as phonological awareness was not examined in this study.

Cardoso-Martins et al. (2009) compared the performance of good and poor readers with Down syndrome on a number of cognitive tasks, including visuospatial measures. Good readers were those who were reading at the level of a typical nine year old or above, and the remainder of the sample formed the poor reader group who all read at the level of a 6-8 year old. The eight good readers performed significantly better than the eleven poor readers on a spatial working memory task, and there was a trend for an advantage on a spatial ability composite score from an IQ test. Spatial working memory was found to be a better predictor of reading accuracy than verbal memory, which suggests that storage and processing of visuospatial information may be more important for reading accuracy. However, the authors highlight the greater variation of scores on the spatial memory task; therefore it may be that that the relative sensitivity of the verbal and visuospatial tasks influenced the results.

As reviewed above, storage and processing of verbal information is impaired in individuals with Down syndrome, however there still appears to be a relationship with reading accuracy. Fidler et al. (2005) found significant correlations between verbal short-term memory and reading accuracy in both their group of children with Down syndrome and the control group of children with learning difficulties, despite the children with Down syndrome performing at a significantly lower level on this task.

Fowler et al. (1995) compared the contribution of verbal and visuospatial short-term memory to word and nonword reading in a group of individuals with Down syndrome. Both memory measures were correlated with both reading measures after controlling for verbal and nonverbal general ability, although the correlations with verbal memory were stronger. After controlling for visual memory, verbal memory accounted for a unique 17% of the variance in reading accuracy, and when verbal memory was controlled, visual memory accounted for an additional 8%. With regards to verbal memory, it appears that a minimum span may be necessary for good decoding, as all participants with decoding skills equivalent to a typically developing 6-7 year old had a digit span of at least three, and participants with decoding skills equivalent to a typically developing nine year old or older had a digit span of at least four. This suggests that both verbal and visual memory are important for learning to read in children with Down syndrome although verbal memory may be more important. Indeed, a minimum level appears to be necessary for decoding.

1.5.1.2. Relationship between reading accuracy, letter-sound knowledge and phonological awareness.

Individuals with Down syndrome have difficulty with phonological awareness tasks in relation to their chronological age, nonverbal ability and reading accuracy. This, despite Cossu et al.'s (1993) claims, does not mean that phonological awareness skills are not relevant to how children with Down syndrome learn to read. Numerous studies have examined the relationship between phonological awareness and reading accuracy in Down syndrome and these will now be reviewed.

Cardoso-Martins and Frith (2001) categorised the individuals with Down syndrome participating in their study as readers or non-readers. There were 46 readers and 47 non-readers; readers were defined as those scoring four or above on a single word reading task. The readers performed significantly better on tasks of initial phoneme detection, letter knowledge, nonverbal cognition and were also younger. The difference in initial phoneme detection was still significant after controlling for age, letter knowledge and nonverbal ability. In their sample of children with Down syndrome Fowler et al. (1995) found that no individual with decoding skills above the level of a 6-7 year old typically developing child scored below chance on a test of phoneme deletion, and although some individuals scored well on phoneme deletion but poorly on decoding, the reverse was not true. Similarly Kennedy and Flynn (2003) found that only three out of their sample of nine children with Down syndrome scored above zero on a nonword reading task, and these were the children who had the highest phonological awareness scores. These studies suggest that phonological awareness skills may be necessary, but not sufficient, for good word and nonword reading for individuals with Down syndrome. However, cut-off scores or median splits of groups result in low statistical power and can lead to children of similar abilities being in separate groups, and it is therefore more informative to investigate correlations between reading skills and phonological awareness.

Roch and Jarrold (2008) compared the relationship between nonword reading and a composite phonological awareness measure of alliteration detection and phoneme deletion in a group of individuals with Down syndrome and a typically developing control group matched on regular word reading. In both groups there was a similar relationship between nonword reading and phonological awareness, which would suggest that individuals with Down syndrome utilise phonological awareness to the same extent as typically developing children whilst decoding. Fletcher and Buckley (2002) found significant correlations between reading accuracy and phonological awareness in a group of 17 individuals with Down syndrome; more specifically, when age and verbal short-term memory were controlled, rime detection and phoneme segmentation correlated with reading accuracy. The participants in both these studies were reading at approximately the level of a seven year old typically developing child. These were, therefore, relatively high-achieving children, and as such the results from these studies may not necessarily generalise to all individuals with Down syndrome.

There are various aspects of phonological awareness and individuals with Down syndrome do not perform equivalently on measures of these different skills. Gombert (2002) examined the relationship between different phonological awareness tasks and reading accuracy in a group of 11 individuals with Down syndrome aged 10-20 years and a group of 11 typically developing children aged 6-8 years matched on reading accuracy. Phoneme counting and phoneme deletion correlated with reading accuracy to a similar extent in the children with Down syndrome and the typically developing children. However, whereas an alliteration oddity task was moderately correlated with reading accuracy in the individuals with Down syndrome, it was negligibly correlated with reading accuracy in the typically developing group. Kennedy and Flynn (2003) also found that alliteration matching was significantly correlated with reading accuracy for participants with Down syndrome, however rime detection, phoneme isolation and phoneme blending were not. Fowler et al. (1995) examined the correlation between phoneme deletion, a relatively complex task of phoneme awareness, reading accuracy and nonword reading; phoneme deletion accounted for 36% of variance in a reading accuracy task and 49% of the variance in a nonword reading task. Therefore phonological awareness is related to reading accuracy in individuals with Down syndrome, and tasks involving the initial sounds of words may be particularly important. However methodological concerns mean that these findings must be treated as preliminary. Kennedy and Flynn provide the raw scores for each of their participants and many of the children did not score above chance on any of the tasks and many of the children scored zero on the phoneme isolation task. Neither Kennedy and Flynn nor Fowler et al. included a control group and both Kennedy and Flynn’s and Gombert’s study used limited sample sizes, eleven and nine respectively.

Boudreau (2002) included a larger sample of children with Down syndrome along with a typically developing control group matched on nonverbal ability and found that in contrast to Kennedy and Flynn (2003), blending was significantly correlated with reading accuracy for the children with Down syndrome, whereas for the typically developing children alliteration oddity was significantly correlated with reading accuracy. No phonological awareness measure correlated with decoding in the individuals with Down syndrome, whereas blending was significantly correlated with decoding in the typically developing group. However decoding was at floor in both groups, and therefore correlations involving this task must be interpreted with caution. Snowling et al. (2002) included a control group matched on reading accuracy meaning that any differences in correlations cannot be due to the different reading levels of the two groups of children. Similar correlations were found between a measure of phoneme detection, which included initial and final phonemes, and reading accuracy in individuals with Down syndrome and the typically developing children.

Letter-sound knowledge is important for acquiring the alphabetic principle that in turn is important for learning to read, more specifically to decode. Kennedy and Flynn (2003) found that the correlation between letter-sound knowledge and reading accuracy was strong for individuals with Down syndrome, and the relationship between letter-name knowledge and reading accuracy was moderate. However this was with a small sample and two other studies with larger groups of children suggest that the relationship between letter-sound knowledge and reading accuracy may not be significant. Snowling et al. (2002) found that letter-name but not letter-sound knowledge was correlated with reading accuracy for individuals with Down syndrome, whereas for typically developing children letter-sound knowledge was strongly correlated with reading accuracy. Boudreau (2002) also found that for children with Down syndrome, letter-name knowledge was significantly correlated with reading accuracy, whereas for the typically developing group, both letter-name and letter-sound knowledge were significantly correlated with reading accuracy. In both these studies, the groups of children with Down syndrome had similar levels of letter-sound knowledge to their control groups, but the lack of a relationship with reading accuracy suggests that they have difficulty applying this knowledge to reading. Conversely, letter-name knowledge was related to reading accuracy in both studies; Snowling et al. (2002) suggests that this relationship may occur not because of a reason associated with the alphabetic principle but rather because both skills involve learning associations between visual referents and their names.

1.5.1.3. Relationship between reading accuracy and broader oral language skills.

With regards to factors affecting reading accuracy in children with Down syndrome, phonological awareness has been most researched. Although phoneme awareness contributes to reading accuracy, this is a weaker relationship than would be expected based on typical reading development, therefore the relationship between reading accuracy and broader oral language has also been the focus of research.

Some studies, although not specifically focussed on oral language skills, have administered language tasks as part of their test battery. As discussed, Fidler et al. (2005) investigated the effect of visual processing on reading accuracy in individuals with Down syndrome and individuals with moderate learning difficulties, but a receptive vocabulary measure was also administered. This correlated with reading accuracy for both groups, despite receptive vocabulary being at a significantly lower level in the Down syndrome group.

Within the oral language domain, individuals with Down syndrome show relative strengths on receptive compared to expressive tasks and also in vocabulary compared to grammar. Therefore it is important that a range of tests are administered to gain a comprehensive understanding of the relationship between oral language and reading accuracy.

Cardoso-Martins et al. (2009) administered a range of language tasks to individuals with Down syndrome, who were divided into good and poor readers. The good readers performed significantly better than the poor readers on a receptive syntax task, and there was also a trend for higher scores on receptive vocabulary and expressive morphology tasks.

Due to the problems associated with median splits, it is important to look at correlations between reading accuracy and oral language. Boudreau (2002) administered such tasks to a group of children with Down syndrome and a typically developing control group matched for nonverbal ability. The oral language tasks included receptive vocabulary, receptive grammar and MLU from a conversational language sample. A narrative task was also administered to produce measures of narrative understanding and production. For the typically developing children, the only language measure which significantly correlated with reading accuracy was narrative production. For the individuals with Down syndrome reading accuracy significantly correlated with receptive grammar and receptive vocabulary. However chronological age was the strongest correlate of reading accuracy and when this was controlled for, the relationships with oral language in both groups were no longer significant. Chronological age is often not an important predictor of ability in individuals with Down syndrome. In this case, as the children with Down syndrome were significantly older than the typically developing children, they are likely to have had more reading instruction and it could be that this then takes over as a powerful predictor of reading accuracy.

The relationship between nonword repetition and reading accuracy has also been examined in children with Down syndrome. Laws (1998) found a significant correlation between nonword repetition and reading accuracy, after controlling for chronological age, nonverbal ability and word repetition, which was included to act as a proxy for speech and perceptual difficulties which may affect nonword repetition. However, the reading accuracy measure in these studies also included letter identification, and it would be more informative to have separate measures of these skills.

1.5.1.4. Longitudinal studies of reading accuracy.

There is evidence for the involvement of a number of skills in the development of reading accuracy in children with Down syndrome. It appears that visual processing and memory, verbal memory, oral language and phoneme awareness may all correlate with the development of reading accuracy. However the research reviewed thus far has been concurrent and consequently it is difficult to establish the direction of the relationships, for example it could be that children with Down syndrome develop better oral language skills as a result of good reading accuracy skills, or that pre-existing good oral language skills help these children to become better readers, or indeed both. There have been a small number of longitudinal studies focussing on the development of reading accuracy, and these can help clarify these relationships.

Cupples and Iacono (2000) investigated the longitudinal relationship between phonological awareness and reading accuracy with 19 children with Down syndrome aged 6-10 years, at two time-points which were approximately nine months apart. Regression analyses were conducted to examine the predictors of reading accuracy and nonword reading. Chronological age, verbal short-term memory and receptive vocabulary were always entered at the first step; however the correlations of these variables with reading accuracy or their independent contributions to outcome variables in the regression are unfortunately not reported. Initial phoneme segmentation ability explained significant additional variance in nonword but not word reading after the autoregressors were controlled for. However, the mean phoneme segmentation scores were close to floor at both time points and several children were reported to not understand the task.

Roch and Jarrold (in press) conducted a follow-up study of 12 of the adolescents and adults with Down syndrome who had taken part in Roch and Jarrold (2008) four years previously. The aim was to examine whether there were any changes in the use of the visual versus phonological route to reading, and to investigate the longitudinal associations between nonword reading, irregular word reading and phonological awareness. To briefly revisit the Time 1 results, phonological awareness was strongly correlated with nonword reading whereas nonword reading and irregular word reading were not significantly correlated with each other. In contrast to these results, at the follow-up phonological awareness was no longer strongly correlated with nonword reading whereas the correlation between irregular and nonword reading was now significant. In regression analyses controlling for the autoregressor, nonword reading did not predict irregular word reading. In contrast irregular word reading did predict later nonword reading, although phonological awareness did not. The authors suggest that with developing word recognition skill, as indexed by irregular word reading, individuals with Down syndrome adopt a different strategy to reading nonwords, namely by the increasing use of visual analogies.

The above studies have concentrated on phonological awareness and different reading outcomes. As illustrated by concurrent studies, skills such as memory and oral language may also influence reading accuracy. One study to include a broader battery of tests is that of Kay-Raining Bird, Cleave and McConnell (2000). They conducted a longitudinal study over four and a half years with 12 children with Down syndrome, who were aged 6-11 years at the outset of the study. There were three time points, with a lag of three years between the first two time-points and 18 months between the second and third time-point. The children were assessed on a range of oral language, reading, memory and phonological awareness tasks at all time-points. Over the course of the study, there were significant improvements in reading accuracy, nonword reading and rhyme generation, but not in phoneme segmentation and deletion, for which the scores were generally low. At every time-point, reading accuracy was better than nonword reading, which the authors suggested shows continued reliance on a visual reading strategy. In terms of the relationship between tasks, after controlling for age and nonverbal ability, the highest correlations for endpoint nonword reading were with initial phoneme segmentation and digit span; however the correlation with phoneme segmentation disappeared after controlling for digit span but not vice-versa. The highest correlations for Time 3 reading accuracy were with initial digit span, phoneme segmentation and phoneme deletion. Kay-Raining Bird et al. also included measures of receptive vocabulary and MLU, and the correlations between these and endpoint reading skills were low to moderate. However, the descriptive scores for the two language measures are not reported, so the correlations cannot be interpreted in the light of the distribution of scores. Given the small sample size and low scores on key measures, including nonword reading and phoneme segmentation, these results must be considered as preliminary.

Laws and Gunn (2002) examined the development of reading, oral language and cognition in a group of 30 individuals with Down syndrome over a period of five years. The individuals who took part were aged 10-24 years at the end of study. They were assessed on receptive language, nonverbal ability, nonword repetition, letter knowledge and reading accuracy at both time points. Hearing thresholds, MLU, alliteration matching and rime matching were also assessed at the second time point. The sample was divided into readers, defined as being able to read at least one word at Time 2, and non-readers. The readers were significantly older than the non-readers and had better nonverbal ability, receptive grammar and MLU. Furthermore, significantly more individuals in the reader group performed above chance on the rime and alliteration matching tasks. There were also group differences in the same direction for nonword repetition and receptive vocabulary, but these were not significant once hearing ability was controlled for. Over the course of the study, five individuals began to read; these individuals had significantly better nonverbal ability and better language skills than those who remained non-readers at the second time-point. The criterion for being a reader was rather conservative and unsurprisingly the range of scores within this group was large, but the between-group analyses do not capture this. Correlations were also conducted and initial receptive vocabulary, receptive grammar and letter-name knowledge were significantly correlated with later reading accuracy.

The longitudinal studies above did not include control groups, however it is important to use a control group to identify similarities and differences with typical development. Over a period of two years, Byrne, MacDonald and Buckley (2002) monitored the progress of 24 children with Down syndrome aged 4-12 years, 21 reading accuracy matched children aged 4-9 years and 32 average readers aged 4-9 years. The average readers were recruited from the same classroom as the children with Down syndrome, but were significantly younger and the reading accuracy matched control group showed delayed reading and language skills relative to their chronological age. The children were assessed on various skills including reading accuracy, verbal memory, visual memory, receptive vocabulary and receptive grammar. Over the two years, children with Down syndrome made significant progress in reading accuracy, although this was significantly slower than the average readers. Overall, children with Down syndrome showed relatively advanced reading accuracy compared to their language ability. The children with Down syndrome and reading accuracy matched children showed generally stronger correlations between reading accuracy and language than the average reader group, and when controlling for age, the correlation between reading accuracy and grammar was greater than that between reading accuracy and vocabulary, which was not significant. Furthermore, when age was controlled, the strongest correlations with reading accuracy for the children with Down syndrome were with both visual and verbal memory. Although typically developing control groups were used in this study, the average reader group were not matched on any variable and although the other group were matched on reading accuracy, they were considered to have delayed reading and therefore may not represent reading in typical development.

Hulme et al. (in press) compared a large group of individuals with Down syndrome with a typically developing group of children matched at the same level of reading accuracy at the beginning of the study. Forty-nine children and adolescents with Down syndrome with a mean age of 10;04 and 61 typically developing children with a mean age of 6;05 took part at three time points, each separated by approximately 12 months. Tasks of nonverbal ability, reading accuracy, vocabulary, letter-sound knowledge and phonological awareness were administered. As found in Byrne et al. (2002), the children with Down syndrome made significantly less progress on reading accuracy than the typically developing children. Path models were used to investigate whether earlier phoneme awareness, vocabulary and letter-sound knowledge predicted reading accuracy after controlling for the autoregressor. In the individuals with Down syndrome, no other variable predicted reading accuracy after the autoregressor, however in the typically developing group earlier phoneme awareness was found to predict reading accuracy development. Due to the strong autoregressive effect of reading accuracy for the individuals with Down syndrome, there was little variance remaining which the other measures could account for. Therefore it may be of interest to examine the predictors of reading accuracy at Time 1. Phoneme awareness was a predictor of reading accuracy in the typically developing children but not vocabulary whereas the reverse was true for the children with Down syndrome. Letter-sound knowledge did not predict concurrent reading accuracy in either group.

1.5.1.5. Training studies to improve reading accuracy.

There is evidence for a relationship between phoneme awareness and the development of reading accuracy in individuals with Down syndrome, although this is weaker than in typical development. As phonological awareness is often a difficulty for individuals with Down syndrome, if this was improved then effects may feasibly generalise to reading accuracy. This provides a rationale for training studies which target phonological awareness. Training studies are useful ways of establishing causation and, in this case, testing whether phonological awareness training can indeed improve phonological awareness skills and subsequently improve reading accuracy in children with Down syndrome.

Cupples and Iacono (2002) contrasted a word analysis intervention, which incorporated phonological awareness, with a whole-word reading intervention with three and four children with Down syndrome aged between eight and twelve years in each condition, respectively. Whilst group statistics were not carried out due to the small sample size, examination of individual's scores suggest that the two conditions had an equal effect on taught words, but only children who received the word analysis training improved on untaught words. This suggests that teaching children with Down syndrome analytic techniques for reading may enable them to generalise these skills to novel words.

A successful reading accuracy intervention with struggling readers who do not have Down syndrome is Hatcher et al.'s (1994) programme which explicitly linked phonological awareness and reading. This approach has also been adopted with children with Down syndrome. Cologon, Cupples and Wyver (2011) adapted this intervention programme for use with seven children with Down syndrome aged 2-10 years, selected to have little or no decoding skill. Compared to a control period when children received normal instruction, the intervention had a significant effect on progress in phoneme blending, phoneme segmentation, reading accuracy (trained and untrained words), nonword reading and letter-sound knowledge. Furthermore these gains were maintained at a six month follow-up assessment. However, performance on phoneme segmentation and untaught words, which were from the same rime families as the trained words, were still low after training. Four of the children in this study were not yet in school and these children made less progress than the older children, suggesting that perhaps with a larger sample of older children, gains would have been greater. However this intervention does demonstrate that children with Down syndrome may benefit from phonics instruction from the very beginning of reading instruction.

Baylis and Snowling (in press) also carried out an intervention based on Hatcher et al. (1994) but focussed on onsets and rimes to minimise the demands on working memory. Ten children with Down syndrome aged 9-14 years took part and they all read at the level of a 6-7 year old typically developing child. From baseline to an immediate post-test, significant gains were seen on tasks of reading accuracy, letter-sound knowledge and phoneme awareness compared to a control period, and these gains were maintained three months later. Despite being trained, rime awareness did not significantly improve. As children with Down syndrome have been found to have specific difficulties with rime awareness, it may be that an intervention targeted at the phoneme level may be even more effective. The control period in this study had a very short duration of two weeks whereas the intervention period was much longer, which makes gains in this time frame more likely.

For interventions to have maximum application to settings outside of research, it is important that individuals other than the researchers can deliver the programme successfully. Therefore Goetz et al. (2008) designed an intervention to be delivered daily and administered by teaching assistants. The format again followed that of Hatcher et al. (1994). Fourteen children with Down syndrome aged 8-14 years took part and they were divided into two groups, an intervention group and a waiting control group, so the potential benefits of the intervention could be compared against normal classroom instruction. The intervention resulted in improvements in reading accuracy, letter knowledge and alliteration matching on immediate and delayed post-tests, and suggests that intensive phoneme awareness and reading intervention can be successful in the short and long-term. Gains on nonword reading were not significant despite having a moderate to large effect size, and it is likely that large variability in scores and small sample size contributed to this. It must also be noted that not all phonological skills improved due to the intervention, for example final phoneme matching was below chance at every time-point.

Lemons and Fuch (2010a) carried out a very intensive reading accuracy and phonological awareness intervention with 24 individuals with DS, aged 7-16 years. This study aimed to examine differential responses to intervention therefore outcome measures of taught letter-sound knowledge, taught sight words, taught regular words and nonwords were administered every three days. There were significant improvements on the measures of taught letter-sound knowledge, taught sight words and taught regular words and there was a trend for growth on the nonword reading task. However not all children benefited from the intervention; eight children were classed as non-responders for regular words and for nonwords, there were 15 non-responders. Children who were better readers at the outset and who had good phonological skills were more likely to benefit from the intervention.

In a review of the literature on reading interventions for children with Down syndrome, Burgoyne (2009) concluded that interventions which trained phonological awareness in the context of letter-sound knowledge were effective for children with Down syndrome, although the studies described to date generally used small sample sizes, short durations and did not benefit all individuals with Down syndrome (Lemons & Fuch, 2010a; 2010b). Oral language is also an important predictor of reading accuracy for children with Down syndrome and Burgoyne suggested that this population of children may benefit particularly from a combined phonological awareness and oral language intervention.

Burgoyne et al. (in press) conducted a randomised controlled trial designed to address the issues outlined above, with a combined phonological awareness and oral language programme. A large sample of 57 children with Down syndrome attending mainstream primary schools were randomly allocated to an intervention group who received 40 weeks of daily intervention or a waiting-control group who received 20 weeks of daily intervention. After the first 20 weeks, the intervention group performed significantly better than the waiting-control group on tasks of reading accuracy, letter-sound knowledge, phoneme blending and taught vocabulary. After the second block of 20 weeks, when the waiting-control group started receiving intervention, only the difference in reading accuracy remained. There was a large degree of variability within the sample and growth in reading accuracy was predicted by age (with younger children showing more growth), intervention attendance and receptive language, a composite of vocabulary and grammar. Randomised controlled trials are considered the gold standard when evaluating treatment programmes, and this stringent test demonstrated that combined training in reading, phonological awareness and oral language produced gains for children with Down syndrome. However even in this intensive intervention, gains were limited to taught skills and did not extend to non-taught oral language and nonword reading.

1.5.1.6. Summary: Predictors of reading accuracy in Down syndrome.

Nonword reading is poorer than reading accuracy in individuals with Down syndrome, which suggests a difficulty in the acquisition and/or application of phoneme awareness and letter-sound knowledge. Therefore these tasks would be expected to have a weaker relationship with reading accuracy than in typical development. Indeed this appears to be the case. There is converging evidence that suggests although phoneme awareness does have a role to play in reading accuracy for individuals with Down syndrome, this is to a lesser extent than for typically developing children and letter-sound knowledge seems not to correlate significantly with reading.

From the small number of concurrent correlational studies in this area, it appears that oral language is concurrently related to reading accuracy in individuals with Down syndrome, and this relationship may be stronger than in typically developing children (Boudreau, 2002). However, it is important to compare performance to reading accuracy-matched control groups as different factors may influence reading accuracy at different stages of development and studies that employ this design are currently lacking.

There are few longitudinal studies which focus on the development of reading accuracy in Down syndrome and small sample sizes, short lapses between time points and the lack of well-matched typically developing control groups makes it difficult to draw strong conclusions about reading development in Down syndrome and how it may differ from typical development. Even when these issues are addressed, such as in Hulme et al. (in press), the relative lack of progress in reading accuracy can make it difficult to find significant predictors of growth.

Despite these issues, reading accuracy appears to be influenced longitudinally by verbal and visual memory, oral language and phonological awareness. As would be predicted from the concurrent studies, phoneme awareness has a relatively small role to play in reading accuracy over time, and there is some indication that vocabulary may be more important for initial reading accuracy skills. Non-phonological oral language has tended to be assessed in longitudinal studies by vocabulary tasks and there is an outstanding need to examine the contribution of a wider range of oral language skills to reading accuracy development.

As a natural progression from the research investigating the relationship between phonological awareness and reading accuracy in children with Down syndrome and the known importance of phoneme awareness in typical reading development, there are a growing number of studies evaluating reading accuracy interventions in Down syndrome. Training phonological awareness, letter-sound knowledge and new vocabulary results in improvements of the directly taught skills. Benefits have also been found to generalise to reading untaught words and there are mixed findings as to whether this generalisation extends to nonword reading. This may be due to difficulty in combining phonological awareness and letter-sound knowledge to acquire the alphabetic principle, which converges on findings from concurrent and longitudinal studies.

1.5.2. Predictors of Reading Comprehension in Down Syndrome

1.5.2.1. The simple view of reading.

The simple view of reading highlights the importance of reading accuracy and oral language to reading comprehension (Gough & Tunmer, 1986). To look at the contribution of decoding to reading comprehension Fowler et al. (1995) categorised 33 individuals with Down syndrome into four groups on the basis of their decoding skill. This classification predicted performance on tasks of reading accuracy and comprehension but most interestingly reading accuracy was significantly better than comprehension and this gap increased as decoding skill increased, i.e. as reading accuracy improves, comprehension falls further behind, which suggests some other limiting factor. Considering the simple view of reading, it is logical to suggest that this limiting factor may be language.

There have not been many correlational studies with children with Down syndrome that have included a measure of reading comprehension. However in Boudreau (2002), reading comprehension and various measures of spoken language were assessed concurrently. It was found that reading comprehension correlated with measures of receptive vocabulary, receptive grammar, MLU and narrative skills. When age was controlled for, MLU was the only language measure which remained significantly related to reading comprehension. The typically developing control group were matched on nonverbal ability and had an average age of 4;05. Due to their young age, they performed very poorly on the reading comprehension task making it difficult to make any meaningful comparisons between the two groups.

It is informative to investigate whether reading accuracy and oral language are differentially related to reading comprehension in typical development and Down syndrome when children have equivalent levels of reading comprehension. Roch and Levorato (2009) used such a design to investigate the contribution of listening comprehension along with word and nonword reading and speed to reading comprehension. A group of individuals with Down syndrome aged 11-18 years were pairwise matched on reading comprehension to a group of typically developing children aged 6-7 years, although the children with Down syndrome had better reading accuracy skills. For the typically developing children, listening comprehension and word reading speed, used as this is a more sensitive measure than accuracy due to Italian's regular orthography, explained unique variance in reading comprehension. However for individuals with Down syndrome, only listening comprehension accounted for significant variance in reading comprehension. This suggests that poor listening comprehension, in the presence of good reading accuracy, may limit reading comprehension in Down syndrome. Levorato, Roch and Beltrame (2009) found that the performance of individuals with Down syndrome on this measure of listening comprehension was predicted by receptive vocabulary and receptive syntax. This suggests that these lower-level oral-language skills may constrain reading comprehension indirectly through listening comprehension.

In Roch and Levorato (2009) the scores of the speeded word reading in the group of children with Down syndrome were close to ceiling and had little variance, which may have limited the potential for a relationship with reading comprehension. However Nash & Heath (2011) also found that language, in this study receptive vocabulary, was more highly correlated with reading comprehension in children with Down syndrome than a group of typically developing children with the same level of reading comprehension. Furthermore vocabulary remained significantly correlated with reading comprehension after reading accuracy had been controlled for. This study also included a group of children at the same reading accuracy level as the children with Down syndrome, and there was also a strong correlation between reading comprehension and vocabulary in this group. Therefore it was argued that the greater contribution of oral language to reading comprehension seen in the group of children with

Down syndrome in Roch and Levorato was the result of higher levels of reading accuracy relative to the typically developing group.

1.5.2.2. Relationship between inference making and reading comprehension.

When reading a text, it is often necessary to go beyond the information explicitly given in the text and make an inference. Very few studies have looked at inference making in children with Down syndrome, but there is some evidence for difficulty in this domain.

Groen et al. (2006) reported the performance of KS, a girl with Down syndrome who had exceptional reading skills, on two tasks of reading comprehension: the Neale Analysis of Reading (NARA II; Neale, 1989) and Wechsler Objective Reading Dimensions Test of Reading Comprehension (WORD; Wechsler, 1990). For both tasks children read passages and answer questions. However, on the WORD the questions tend to be literal, whereas on the NARA more inferences are required (Bowyer-Crane & Snowling, 2005). KS performed worse on the NARA compared to the WORD leading the authors to suggest that she had a particular weakness in generating inferences.

The comparison between questions which require inferences or literal information was explored in a larger group of good readers with Down syndrome by Nash and Heath (2011). Four stories were designed which were followed by an equal number of literal and inferential questions. Children with Down syndrome were compared to typically developing children at the same reading accuracy level, typically developing children at the same reading comprehension level and poor comprehenders who were also matched for reading comprehension. The children with Down syndrome performed equivalently to the two groups of children matched on reading comprehension on literal questions, but scored significantly below these two groups on inferential questions. Therefore there is a clear discrepancy between literal and inferential questions in Down syndrome, which was also present to a lesser degree for the children with reading comprehension impairment. This supports Groen et al.'s (2006) case study and suggests that children with Down syndrome may have a specific problem with inference-making and it is possible that this contributes to their reading comprehension difficulties.

1.5.2.3. Relationship between working memory and reading comprehension.

Verbal working memory is necessary for successful reading comprehension and children with Down syndrome have well documented difficulties with working memory. Therefore it is possible that this could limit their reading comprehension skills. The contribution of working memory to reading comprehension in children with Down syndrome and how this compares to typically developing children was explored by Levorato, Roch and Florit (2011). The two groups of children were matched on reading comprehension, and also performed similarly on a working memory task. Working memory contributed unique variance to reading comprehension, after sentence comprehension and a short-term memory task were controlled for. Furthermore the relationship between working memory and reading comprehension was similar in the two groups. In comparison, Nash and Heath (2011) found a stronger relationship between reading comprehension and working memory in children with Down syndrome than a group of typically developing children at the same level of reading comprehension; although this relationship was similar once reading accuracy was controlled for.

1.5.2.4. Longitudinal studies of reading comprehension.

The longitudinal studies with children with Down syndrome tend to either not include reading comprehension or not treat it as an outcome measure. The few studies that have, have noted poor progress, at a slower rate than for reading accuracy (Byrne et al., 2002; Laws & Gunn, 2002).

The only longitudinal study to specifically look at reading comprehension development in children with Down syndrome was carried out by Roch, Florit and Levorato (2011). Ten individuals with Down syndrome aged 11-19 years participated, and their reading comprehension, listening comprehension and reading accuracy skills were assessed at two time points, 12 months apart. A regression analysis was conducted to investigate which initial skills predicted later reading comprehension. After the autoregressor of reading comprehension had been entered, reading speed contributed 8% of variance, which was not significant, and listening comprehension contributed 32% of variance, a significant proportion. This confirms the pattern seen in the concurrent study by the same authors (Roch & Levorato,

2009) of listening comprehension playing a larger role in reading comprehension for children with Down syndrome than reading speed. However due to the three-step regression analysis on data from only 10 participants, these results need replicating with a larger sample size.

1.5.2.5. Summary: Predictors of reading comprehension in Down syndrome.

Children with Down syndrome have difficulties with reading comprehension in comparison to their reading accuracy skills. Considering the simple view of reading as a framework and weaknesses in the verbal domain, it is likely that oral language may limit reading comprehension. Indeed for children with Down syndrome who do reach good levels of reading accuracy, this appears to be the case. Higher-level processes, including working memory and inference-making, are also related to reading comprehension in Down syndrome. Again both these domains appear to cause difficulty for children with Down syndrome, which may then limit reading comprehension.

There has thus far only been one longitudinal study to specifically look at the predictors of reading comprehension in children with Down syndrome. The results confirmed the greater contribution of oral language compared to reading accuracy seen in concurrent studies. However there is a pressing need for longitudinal studies with typically developing control groups, larger sample sizes and a larger test battery of possible predictors to confirm and extend these findings.

1.5.3. The Impact of Reading Accuracy on Oral Language in Down Syndrome

Children with Down syndrome have oral language and verbal memory difficulties; therefore the use of visual support has often been encouraged in their education. Buckley (1995) proposed that seeing the written form of words helps the spoken language development of children with Down syndrome. In support of this, some case studies suggest that teaching young children with Down syndrome to read promotes language development, particularly in the production of words or sentences which are first introduced in their written form (de Graaf, 1993; Duffen, 1976).

The proposal that reading accuracy may promote oral language development was examined by Laws, Buckley, Bird, MacDonald and Broadley (1995). Fourteen children with

Down syndrome aged 8-14 years took part in a longitudinal study and after four years half of the children were able to read at least one word on a reading task and so were classified as readers. Nonverbal ability, receptive vocabulary, receptive grammar and memory were assessed at both time points. It was found that readers made greater progress than non-readers on memory measures, and there was a trend in the same direction on the vocabulary and grammar tasks, which supports earlier case studies. However the group of children who could read had more advanced language skills at the first time point. Furthermore, all the children who could not read were in special education and those who could read were mostly in mainstream education, which has been found to lead to greater progress in spoken language (Buckley et al., 2006).

The relationship between earlier reading accuracy and later oral language was examined in Laws and Gunn's (2002) longitudinal study of individuals with Down syndrome. Reading accuracy was not correlated with receptive language five years later but was significantly related to later MLU and phonological awareness. Unfortunately as MLU and phonological awareness were not assessed at Time 1, the reciprocal relationship of these skills with reading accuracy and the effect of the autoregressor cannot be assessed.

Studies have also examined the relationship between earlier reading and later phoneme awareness whilst controlling for the autoregressor. Cupples and Iacono (2002) found that initial phoneme segmentation was the best predictor of later phoneme segmentation, and neither word nor nonword reading added unique variance. In comparison Hulme et al. (in press) found that the pathway from earlier reading accuracy to later phoneme awareness was significant, along with the autoregressive pathway. Furthermore the strength of this relationship was similar in the group of children with Down syndrome and typically developing children. Therefore it appears that reading may influence later phoneme awareness, but results are contradictory as to whether this holds over and above the autoregressor.

1.6. Reading and Language Skills in Down Syndrome: Summary, Conclusions and Thesis Aims

This chapter has outlined our current understanding of reading development in typically developing children to provide a framework in which to consider reading in children with Down syndrome. The level of reading accuracy, reading comprehension, oral language and related skills in individuals with Down syndrome has been discussed and evidence for causal relationships from concurrent, longitudinal and intervention studies has been reviewed.

Reading accuracy development appears to differ in children with Down syndrome compared to typically developing children, and this is likely due to the well-documented weaknesses in phonological awareness. There are two lines of evidence to support this. Firstly there is a discrepancy between nonword reading and word recognition, indicating difficulties in decoding for which phoneme awareness is necessary. Secondly, studies investigating the relationship between phonological awareness and reading accuracy in children with Down syndrome indicate that although phoneme awareness does appear to impact upon reading skills, this is to a lesser extent than in typical development. As phoneme awareness is below reading accuracy levels in individuals with Down syndrome, other skills may be compensating and promoting good word recognition skills and it appears that oral language may have a particularly important role to play, although studies with large oral language test batteries and control groups matched on reading accuracy are lacking.

Children with Down syndrome are often able to reach relatively high levels of reading accuracy. With this progress, it is apparent that reading comprehension is a weakness. The contributing skills to reading comprehension in typical development, such as oral language, working memory and inference-making, are also impaired in children with Down syndrome. This, and the presence of relationships between these skills and reading comprehension, suggests that they may act as a limiting factor in reading comprehension development once reading accuracy has reached an adequate level.

In contrast to reading, oral language is a relative weakness for children with Down syndrome. It is of particular interest that reading accuracy has been suggested to promote oral language development as it can provide visual support. There is some evidence to support this, but more methodologically robust longitudinal studies with typically developing control groups are needed.

This thesis aimed to explore reading and oral language development in children with Down syndrome and more specifically investigated the interaction of these two domains in comparison to typically developing children at the same level of reading ability. Three studies were conducted, which utilised two complementary methodologies: a longitudinal study and two word learning experiments.

The longitudinal study explored the contribution of a range of cognitive tasks to the outcome measures of reading accuracy, reading comprehension and oral language in a group of children with Down syndrome compared to typically developing children matched for initial levels of reading accuracy. The aims of this study were to evaluate the contribution of phoneme awareness and a broad battery of oral language tasks to reading accuracy, to consider the simple view of reading in children with Down syndrome and examine any additional contribution from memory, and to investigate the possible benefit from reading accuracy on oral language growth.

The two experiments approached the interaction between written and spoken language in a different manner. Spoken and written word learning paradigms were utilised to investigate the effect of reading accuracy on phonological learning, and the effect of phonology on learning to read new words. The spoken word learning study compared children's ability to learn a novel phonological form and semantic referent with and without the written form (i.e. the orthography) being present during training. The written word (i.e. orthographic) learning study tested the benefit of pre-training the phonology of nonwords before children learnt to read them. In both these studies, a control group of typically developing children at the same level of reading accuracy were included to test for any differences between children with Down syndrome and typical development.

Chapter Two

A Longitudinal Study of Reading and Language Development in Children with Down Syndrome

2.1. Introduction

2.1.1. Overview

The development of reading accuracy in children with Down syndrome is related to a number of different factors, as in typically developing children. However differences with typical development appear to lie in the relative importance of different factors; specifically phoneme awareness appears to have a less important influence on the development of reading accuracy in children with Down syndrome, whereas broader language skills may play a more important role (e.g. Boudreau, 2002; Hulme et al., in press). In order to achieve a comprehensive understanding of how reading accuracy relates to phoneme awareness and broader oral language skills, there are a number of methodological issues that need to be addressed.

Most studies thus far have been concurrent and therefore the direction of the relationship between different skills is unclear. There is a dearth of longitudinal studies which measure phoneme awareness and broader oral language skills at all time points. Furthermore, control groups are often not included or are matched on variables such as nonverbal ability (e.g. Boudreau, 2002). It is important to have a control group matched on reading accuracy so any differences between the groups cannot be due to their stage of reading development. Additionally, language measures are often lacking from studies or only receptive tasks are used. Receptive tasks are typically easier and quicker to administer and remove the requirement for a verbal response, which could be a confounding factor as individuals with Down syndrome often have expressive language difficulties. However in order to gain a comprehensive understanding of the impact of oral language skills on reading accuracy, a wide range of measures need to be administered including expressive language tasks.

To read for meaning is the ultimate goal of reading, and therefore it is important to consider reading comprehension as a separate skill to reading accuracy, especially as there is evidence that these two skills can be dissociated. There are fewer studies focussing on reading comprehension than reading accuracy in children with Down syndrome, although as the standards of literacy achieved by children with Down syndrome increase, their reading comprehension ability is becoming a focus of more studies. It is clear that children with Down syndrome have reading comprehension levels below that expected from their reading accuracy (e.g. Nash & Heath, 2011; Roch & Levorato, 2009). Oral language is a key skill for reading comprehension, and as children with Down syndrome generally have difficulties in this domain, this may be expected to place limits on their comprehension of written material (Roch & Levorato, 2009). Oral language does indeed appear to be more strongly related to reading comprehension in children with Down syndrome, although this may be because children with Down syndrome tend to be at a more advanced level of reading accuracy than their typically developing control groups (Nash & Heath, 2011; Roch & Levorato, 2009; Roch et al., 2011). As with reading accuracy, there is a shortage of longitudinal studies in this area. Currently only one study has specifically investigated longitudinal predictors of reading comprehension in Down syndrome but this had a small sample size and no typically developing control group (Roch et al., 2011).

Two strengths typically observed in the cognitive profile of children with Down syndrome are reading accuracy and visual skills. This has led to the suggestion that reading accuracy could be used to promote oral language development (e.g. Buckley, 1995). This has been addressed by a handful of longitudinal studies but the results are mixed, possibly due to whether the autoregressor is accounted for or not (Cupples & Iacono, 2002; Hulme et al., in press; Laws & Gunn, 2002) and methodological issues such as comparing groups of readers and non-readers with Down syndrome who were not matched for initial oral language level (Laws et al., 1995). Furthermore the possible impact of learning to read on oral language development beyond phoneme awareness for children with Down syndrome has not yet been compared to typically developing children. This is an important omission as the implication is that this relationship is special or unique to individuals with Down syndrome due to their cognitive profile.

2.1.2. Aims of Study 1

There are outstanding questions regarding the interrelationships between reading accuracy, reading comprehension and oral language development in children with Down syndrome and how this compares to typical development. Study 1 aimed to address this with a longitudinal design including tasks to assess all three domains at each time point.

A group of children with Down syndrome and a group of typically developing children matched for initial levels of reading accuracy were assessed three times over a two year period. Reading accuracy, reading comprehension, letter-sound knowledge, phonological skills, grammar, vocabulary, narrative skills, memory and nonverbal ability were assessed. As reading accuracy is typically found to be an 'island of ability' for individuals with Down syndrome, it was expected that the children with Down syndrome would perform significantly worse than the control group on all other measures at the first time-point. At the subsequent time-points it was expected that the typically developing children would make more progress and therefore the two groups would be no longer matched on reading accuracy.

There is clear evidence to suggest that phoneme awareness and letter-sound knowledge are related to reading accuracy to a lesser degree in children with Down syndrome compared to typically developing children. There is some evidence to suggest oral language may be more important than phoneme awareness for reading accuracy in children with Down syndrome, whereas at beginning stages of reading this does not appear to be the case for typically developing children. It was expected that these findings would be replicated, with broader oral language skills being a more important predictor than phoneme awareness for reading accuracy in children with Down syndrome and the opposite pattern being evident for the typically developing children. By including both receptive and expressive measures, the contributions of different aspects of language could be explored.

The relationships between reading comprehension, reading accuracy and oral language were also examined. According to the convergent skills model, at the beginning of literacy development reading accuracy is more strongly related to reading comprehension than oral language but as children's reading accuracy improves, oral language takes over as a stronger predictor of reading comprehension (Vellutino et al., 2007). Therefore at Time 1, both groups of children were at the early stages of reading development and therefore reading accuracy was expected to predict reading comprehension more strongly than oral language. At Time 3, the typically developing children will have progressed in their reading comprehension skill and oral language would therefore also be expected to play a role. However as the children with Down syndrome were not expected to make as much progress in reading comprehension, oral language may be less likely to predict reading comprehension longitudinally.

The impact of reading on oral language was the third area of interest. It was expected that reading accuracy would predict later phonological skills for the typically developing children (Hulme et al., in press; Nation & Hulme, 2011) and children with Down syndrome (Buckley, 1995; Hulme et al., in press). The effect of reading accuracy on broader oral language skills has not been widely investigated in typical development. There is some support for this relationship in children with Down syndrome but there is a need for better controlled longitudinal studies (Laws et al., 1995; Laws & Gunn, 2002). With the exception of Hulme et al. (in press), who examined the effect of reading accuracy on phoneme awareness, no studies have compared the effect of reading accuracy on oral language development in children with Down syndrome to typically developing children. Therefore the effect of reading accuracy on broader oral language skills and the comparison between children with Down syndrome and typical development was to be explored.

2.2. Method

2.2.1. Design

Children with Down syndrome and a control group of typically developing children matched for reading accuracy level at Time 1 were assessed on a battery of general ability, memory, literacy and language tests. There were three time-points of assessment. Time 1 took place between February and July 2009. Time 2 occurred between February and July 2010; the average lapse between Time 1 and Time 2 was 12.26 months for the children with Down syndrome and 12.17 months for the typically developing children. Time 3 took place between January and March 2011. The delay between Time 2 and Time 3 was 10.38 months for the children with Down syndrome and shorter at 8.96 months for the typically developing group. This difference between the two groups was significant, $U = 0.00$, $p < .001$. The discrepancy between the two groups was due to logistical reasons; as testing was scheduled to be completed by the end of the spring term, typically developing children were tested earlier in the year than at previous time-points.

2.2.2. Participants

2.2.2.1. Children with Down syndrome.

Children with Down syndrome were recruited by contacting Down syndrome support groups in Yorkshire and the North-East of England. The criteria for inclusion were that children had to be aged 6-16 years and had to be able to read according to parental report. All children also used speech as their primary mode of communication. The parents of children who had previously taken part in research at the Psychology Department, University of York and who met the criteria above were also contacted. Consent forms were completed by all parents or guardians to allow the children to take part. At the outset of the study 23 children with Down syndrome were involved, but one child moved away from the area before Time 2 and another child did not complete any expressive language tasks at all time-points and also no reading tasks at Time 3 so was excluded from all analyses. Therefore, data from the 21 remaining children are reported (see Table 1 for participant characteristics at Time 1). According to parental questionnaires, 20 children had Trisomy 21 and one parent did not know what form of Down syndrome their child had. At Time 1, 10 children attended mainstream primary schools, one of whom had joint attendance with a special primary school. Eight children attended mainstream secondary schools and three children attended special secondary schools.

Hearing thresholds at 1000Hz and 4000Hz were tested for the children with Down syndrome with a pure tone audiometer, as they are often reported to have hearing impairments (Shott et al., 2001). Data for 15 participants were available, although one of these children had a hearing aid in her left ear and therefore hearing threshold was not tested for this ear. Hearing tests were not completed for the remaining six children for a number of reasons including equipment availability and behavioural issues. According to the Royal National Institute for Deaf People's (RNID) criteria for hearing loss (RNID, n.d.), when thresholds were averaged across both ears, at 1000Hz six children presented with mild hearing

Table 1.

Gender, age and mean reading scores (standard deviations) for the children with Down syndrome and typically developing children (Study 1)

	Children with Down syndrome (n=21)	Typically developing children (n=29)
Gender (male: female)	9:12	12:17
Age	11;05 (2;10)	6;02 (0;10)
Single word reading score	15.95 (11.85)	16.41 (12.64)

loss and one child had moderate hearing loss, and at 4000Hz, two children had mild hearing loss and two children had moderate hearing loss. The average threshold across both ears for 1000Hz was 24.50db (standard deviation of 8.46) and for 4000Hz it was 21.33db (standard deviation of 13.82). This level of hearing is consistent with previous research (e.g. Marcell & Cohen, 1992).

2.2.2.2. Typically developing children.

Typically developing children were recruited from two primary schools in York which serve areas of average to above average socio-economic status. Consent was gained from the headteachers of the schools and consent forms were then sent to the parents of randomly selected children in Reception, Year 1 and Year 2, excluding children who spoke English as a second language or those who had been identified with special educational needs. There were two strands to the assessment of the control group. Firstly all children for whom parental consent was received were given the York Assessment of Reading Comprehension (YARC)-Passage Reading single word reading test (Snowling et al., 2009). Children who matched a participant with Down syndrome on gender and word reading (within four words, except one child who had a discrepancy of eight words from a child with Down syndrome) were then given the full assessment battery. This was to ensure that the typically developing group had a similar word reading raw score (both means and standard deviation) to the group of children with Down syndrome (Facon, Magis, & Belmont,2011). Twenty-nine typically developing children were included so if attrition occurred there would still be at least as many typically developing children as children with Down syndrome. It can be seen from Table 1 that the two groups were matched on word reading scores and this was confirmed by a Mann-Whitney U test (U=317.00, *p*=.97), which conforms to Mervis and Klein-Tasman's (2004) recommendation that if the groups are matched, the p value for between-group differences should be above .50.

2.2.3. Assessment Battery

The assessment battery was largely similar at Time 1 and 2 but was reduced at Time 3, due to concentration on outcome measures and the omission of tests on which the typically developing group would be expected to perform at ceiling. Table 2 shows the content of the assessment battery at each time point. Appendix 1 shows the reliability of all the standardised tests as reported in the manuals.

2.2.3.1. Non-verbal skills (Time 1 and Time 2).

The matrices subtest from the Wechsler Pre-School and Primary Scale of Intelligence III^{UK} (WPPSI-III^{UK}; Wechsler, 2003) or Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) was administered to measure nonverbal abstract reasoning skills. In this test, children are asked to look at an incomplete matrix and choose the missing section from four or five options. For both versions of the tasks, testing was discontinued after four consecutive scores of 0, or four scores of 0 on five consecutive items.

The WASI matrices subtest is suitable for children aged six years or above, and therefore this was used for this age range in the typically developing group. For children aged below six years, the WPPSI-III^{UK} matrices subtest was administered. Most of the children with Down syndrome were administered the WPPSI-III^{UK}, as previous research suggests that individuals with Down syndrome of similar chronological ages to the participants in this study tend to obtain nonverbal age-equivalent scores of 4-5 years (Boudreau, 2002; Chapman et al., 1998; Laws, 2002; Price et al., 2007). Indeed, none of these individuals performed at ceiling on the WPPSI-III^{UK} matrices subtest. Two of the participants had taken part in previous research at the Department of Psychology, and were known to be of a higher nonverbal IQ and therefore the WASI was administered. Due to the use of different tasks, age-equivalent scores were used in the analyses.

Table 2.

The content of the assessment battery at each time point (Study 1)

	Time 1	Time 2	Time 3
Nonverbal skills	Matrices	Matrices	-
Memory	Digit recall	Digit recall	-
	Block recall	Block recall	-
Literacy	Letter-sound knowledge	Letter-sound knowledge	-
	Early word reading	Early word reading	-
	Single word reading	Single word reading	Single word reading
	Passage reading	Passage reading	Passage reading
	Nonword reading	Nonword reading	Nonword reading
Phonological skills	Alliteration matching	Alliteration matching	Alliteration matching
	Nonword repetition	Nonword repetition	-
	-	Phoneme deletion	Phoneme deletion
Vocabulary	Picture naming	Picture naming	Picture naming
	BPVS	BPVS	BPVS
Grammar	Sentence structure	Sentence structure	-
Narrative skills	Narrative	Narrative	Narrative

2.2.3.2. Memory.

2.2.3.2.1. Verbal memory (Time 1 and Time 2).

The digit recall subtest from the Working Memory Test Battery for Children (WMTB-C; Pickering & Gathercole, 2001) was used to measure verbal short-term memory skills. The children heard a list of digits and had to repeat it in the same order. The lists increased in length until the span score had been determined.

2.2.3.2.2. Visuospatial memory (Time 1 and Time 2).

The WMTB-C block recall subtest measured visuospatial memory. The children had to remember the order in which a series of blocks were tapped by the examiner and reproduce the sequence. The sequences increased in length until the span score had been determined. For both the memory tasks the number of correct trials, rather than span score, was used in analyses.

2.2.3.3. Literacy.

2.2.3.3.1. Letter-sound knowledge (Time 1 and Time 2).

The letter-sound knowledge subtest from the YARC Early Reading test battery (Hulme et al., 2009) was used to assess children's knowledge of 32 grapheme-phoneme correspondences. Children were shown all the letters of the alphabet and six digraphs in a set order and asked to produce the sounds that they represent. If the child responded with the letter name then they were reminded to produce the sound.

2.2.3.3.2. Early word reading (Time 1 and Time 2).

The early word reading subtest from the YARC Early Reading test battery was used to assess children's knowledge of 30 high-frequency words such as 'cat' and 'dragon'. The test was discontinued if the child answered 10 consecutive items incorrectly.

2.2.3.3.3. Single word reading (all time points).

The single word reading test consists of 60 words that increase in complexity from simple words such as 'see' to more complex words such as 'pseudonym'. Children were shown all words and asked to read as many as they could.

As described in the 'Participants' section, all typically developing children were given the single word reading test first. If the child read the first 20 items correctly then full credit was given for the early word reading test, as this test was deemed to be below their reading accuracy level. If children with Down syndrome were thought to be good readers either from participation in previous research or parental/teacher report then they were also given the single word reading test first, and if they too read the first 20 items correctly then full credit was given for the early word reading test. To ensure fair comparison across the sample any child with Down syndrome who completed both reading tasks and answered the first 20 items correctly on the single word reading test was also given full credit for the early word reading test regardless of their actual score.

2.2.3.3.4. Passage reading (all time points).

The YARC Passage Reading test was used to obtain scores for reading accuracy and reading comprehension. This test consists of seven passages for children to read aloud and then answer open-ended questions about. Raw scores on this task for both reading accuracy and comprehension were transformed into ability scores.

According to the YARC instruction manual, the typically developing children were administered the passage which corresponded to their single word reading test score and then depending upon their performance, the next passage at either a higher or lower level was administered. Hence each child completed two passages.

This test has not yet been used with children with Down syndrome and therefore all children with Down syndrome were administered the reception level passage and continued through the test until their reading accuracy errors met the discontinuation rule. However the passages used in the data analysis for the children with Down syndrome were calculated in the same way as for the typically developing group, i.e. by reference to their single word reading test scores.

2.2.3.3.5. Nonword reading (all time points).

The Graded Nonword Reading Test (GNWRT; Snowling, Stothard & McLean, 1996) was used to test children's decoding skills. Participants were presented with nonwords which increased in difficulty from 'hast' to 'sloskon', and were asked to read them aloud. There were 20 items in total and five practice trials, and the task was discontinued after six consecutive errors. To increase the difficulty of the tests at Time 2 and Time 3, five extension items were added from the phonetic decoding efficiency subtest of the Test of Word Reading Efficiency (TOWRE; Torgesen, Wagner & Rashotte, 1999). These were bramtich, chimdruff, darlenkert, obsorfelm and pitocrant.

2.2.3.4. Phonological skills.

2.2.3.4.1. Alliteration matching (all time points).

To assess phonological awareness children were given a test of alliteration matching, which was adapted from Carroll (2004). All stimuli were presented to children as spoken words and colour pictures. Children were asked which word out of a choice of two started with the same sound as a target word. There were two practice items and 10 test items. The distracters were matched to the correct answer for global similarity to the target word. The items for this task are shown in Appendix 2.

2.2.3.4.2. Phoneme deletion (Time 2 and Time 3).

A phoneme deletion test from McDougall et al. (1994) was used, which includes 24 items requiring the deletion of initial, medial and final phonemes from nonwords. A practice item was also added. All the items for this task are shown in Appendix 3.

All children were administered the alliteration matching task, and were also administered the phoneme deletion task at Times 2 and 3 if they performed significantly above chance on the alliteration matching test, i.e. answered eight or more correct. If children were not administered the test then this was treated as missing data. The phoneme deletion task was expected to be particularly difficult for children with Down syndrome based on previous research therefore this procedure was used to avoid unnecessary testing.

2.2.3.4.3. Nonword repetition (Time 1 and Time 2).

The Pre-School Repetition Test (PSRep) from the Early Repetition Battery (ERB; Seeff-Gabriel, Chiat, & Roy, 2008) was used to test phonological memory, i.e. how well children can retain strings of speech sounds in memory. Children were asked to repeat 18 words such as ‘police’ and 18 nonwords such as ‘lepeese’. The words and nonwords ranged from one to three syllables in length. To extend the test to include four syllable items, six words (helicopter, macaroni, supermarket, cauliflower, watermelon, caterpillar) and six nonwords were added (celihopter, racamoni, mupersarket, fauliclower, materwelon, patercillar). These nonwords were created in line with the PSRep nonword items so had the same prosodic structure as the words but transposed two consonants. The word repetition test was used to provide information about children's articulation; if children made consistent errors on the word repetition task then this was taken into account on the nonword repetition task. For Time 2, five nonwords were added: yurnimotaiv, pelarfissoel, zornarvoobim, bikeemevorp, fooresolarn. These nonwords were created to be word-like, which was measured using the phonological neighbourhood density of the syllables.

2.2.3.5. Broader oral language skills.

2.2.3.5.1. Expressive vocabulary (all time points).

The WPPSI-III^{UK} picture naming subtest was administered to test children's expressive vocabulary ability. Children had to name a series of 30 pictures ranging from car to thermometer, and the test was discontinued if five consecutive incorrect responses were made. For Time 2 and Time 3, seven items were added from the Expressive One Word Picture Vocabulary Test (EOWPVT; Brownell, 2000). These were saddle, chess, banjo, boomerang, greenhouse, pier and microscope and were chosen to be of a greater difficulty than the items in the WPPSI-III^{UK} picture naming subtest.

2.2.3.5.2. Receptive vocabulary (all time points).

The British Picture Vocabulary Scale II (BPVS II; Dunn, Dunn, Whetton, & Burley, 1997) was used to assess children's receptive vocabulary. The children heard a word and had to choose the corresponding picture from a choice of four. The test was discontinued when eight incorrect responses were made in a block of 12 items.

2.2.3.5.3. Receptive syntax (Time 1 and Time 2).

The Clinical Evaluation of Language Fundamentals-Preschool 2^{UK} (CELF-Preschool 2^{UK}; Wiig, Secord, & Semel, 2006) or the Clinical Evaluation of Language Fundamentals-Fourth UK Edition (CELF-4^{UK}; Semel, Wiig, & Secord, 2006) sentence structure subtest was used to measure receptive syntax. This subtest assessed comprehension of sentences that increased in structural complexity. Children were asked to point to the picture, from a choice of four, which depicted a spoken sentence. All 26 items were administered in the CELF-4^{UK}. There were 22 items in the CELF-Preschool 2^{UK}, which was discontinued after five consecutive scores of zero.

The CELF-4^{UK} is suitable for children aged six or over, and was used for this age range in the typically developing group. Children aged five years were administered the CELF-Preschool 2^{UK}. As grammar is typically a weakness for children with Down syndrome, most individuals were administered the CELF-Preschool 2^{UK}, and none of these individuals approached ceiling on this task. However children with Down syndrome who were known to have relatively good grammatical skills from previous research were administered the CELF-4^{UK}. Due to the use of different tasks, age-equivalent scores were used in the analyses.

2.2.3.5.4. Narrative skills (all time points).

A narrative task was used to obtain MLU and a narrative content score. Language samples from either free play or conversations are often used to calculate MLUs, but it has been suggested that children with Down syndrome have lower MLUs in such contexts compared to when narratives are used (Miles et al., 2006).

The narrative task consisted of five pictures which depicted a sequential story on the topic of a snowy day. Children were asked to tell a story using the pictures and where necessary, non-specific prompts such as ‘what’s happening in the next picture?’ were used. The narratives were audio recorded so they could be transcribed at a later date.

The narratives were transcribed and MLUs calculated based on a set of rules adapted from the Expression, Reception and Recall of Narrative Instrument (ERRNI; Bishop, 2004). Due to intelligibility and expressive language weaknesses typically seen in individuals with Down syndrome, intelligibility and low number of utterances were additional issues. Utterances were excluded from the MLU computation if they contained 50% or more unintelligible words. At Time 1, the number of utterances ranged in the typically developing group from 4 to 17, with a mean of 7. To make the groups comparable, at all time-points only individuals who made at least four utterances were included in the analyses.

The narrative content score was designed to measure how well the children’s narratives reflected the key events in the pictures. Fourteen content units within the story were identified and for each of these children were given a score of 0-3 points for each depending on how much appropriate detail they provided. The scoring guidelines can be seen in Appendix 4.

2.2.4. Procedure

The children with Down syndrome were tested either at home or school and some children had their teaching assistant or parent present depending on school and parental preference. At Time 1 the tests were administered, where possible, in the following order: matrices, picture naming, letter-sound knowledge, early word reading, single word reading, alliteration matching, GNWRT, BPVS, sentence structure, digit recall, block recall, passage reading, narrative. The test order remained largely the same at Time 2. Phoneme deletion was administered after alliteration matching and passage reading was now administered after the single word reading test. The order for Time 2 was followed at Time 3 but omitting the tasks which were no longer included in the test battery. For Time 2 and Time 3, each child was seen 2-3 times and each testing session lasted 20-60 minutes depending on the child's age and concentration. Due to the reduced test battery at Time 3, children were seen once or twice.

The testing of the typically developing children took place in a quiet place within their school. All children were administered the single word reading test first at Time 1. The children selected for further assessment were then seen a further 2-3 times and each session lasted 10-30 minutes, again depending on the child's age and concentration. At Times 2 and 3, most children were seen for two sessions. At each time-point, the tests were carried out in the same order as for the children with Down syndrome.

2.3. Results

The aims of this longitudinal study were three-fold: to examine the relative importance of phoneme awareness and broader oral language skills as predictors of reading accuracy; to evaluate the impact of reading accuracy and oral language skills on reading comprehension; to investigate the effect of reading accuracy on oral language development. To address these aims, the descriptive statistics for each group at each time point will be reported, followed by concurrent correlations and path models focussing on the relationships with reading accuracy and reading comprehension. Longitudinal correlations are then outlined, depicting the relationship between Time1 measures and Time 3 reading accuracy, reading comprehension and oral language. Longitudinal path models were also conducted with reading accuracy and reading comprehension as outcome measures.

An alpha level of .05 was used for all analyses. Raw scores were used, with two exceptions. For the matrices and sentence structure tests, different participants completed slightly different versions according to their age or ability, and therefore age-equivalent scores were used for these tasks.

2.3.1. Descriptive Statistics at Time 1

The mean scores (and standard deviations) for all the tasks at Time 1 are shown in Table 3. All data were complete with the exception of one child who did not complete the alliteration matching task and four children with Down syndrome for whom MLU was not calculated as their total number of utterances was below four (see section 2.2.3.5.4. for details).

To examine the distribution of scores within the groups the Shapiro-Wilk test was used and histograms, along with measures of skew and kurtosis, were examined. A number of measures for both groups were not normally distributed. For the matrices task, there was a positive skew for the children with Down syndrome as most children scored towards the lower end of the distribution. Both groups tended to score highly on the early word reading test, leading to negative skews. Approximately half the children with Down syndrome did not score on the nonword reading task, and half of the typically developing group scored under five leading to high scores of kurtosis. For the letter-sound knowledge task the scores in both groups were negatively skewed. There was evidence of a ceiling effect for the typically developing children on the alliteration matching task, with over half of the children at the maximum score. For the nonword repetition task, the typically developing children's scores were clustered around the higher end of the distribution, whereas the opposite was true for the children with Down syndrome. On the sentence structure task, the typically developing children tended to either score quite low or high with few children scoring in between, and the scores were generally quite low with little variation for children with Down syndrome. When using parametric statistics it is important to take into consideration the nature of these

Table 3.

Scores of the children with Down syndrome and typically developing children on all Time 1 measures (Study 1)

	Scores of the children with Down syndrome		Scores of the typically developing children		
	Mean (standard deviation)	Range	Mean (standard deviation)	Range	Between-group differences
Matrices (age-equivalent score)	4;08 (1;02)	4;00-8;01	6;07 (1;06)	4;00-9;06	U=538.00, *p*<.001
Early word reading (max.30)	20.57 (10.29)	0-30	20.48 (9.80)	1-30	U=347.00, *p*=.589
Single word reading (max.60)	16.67 (11.65)	0-34	16.41 (12.64)	0-38	t(49)=-.13, *p*=.896
Passage reading accuracy	28.95 (13.65)	4-45	35.79 (16.28)	4-63	t(48)=-1.57, *p*=.124
Passage reading comprehension	18.05 (13.17)	0-37	37.86 (13.89)	10-55	t(49)=-4.62, *p*<001
Nonword reading (max. 20)	3.14 (4.60)	0-13	8.90 (7.24)	0-19	U=454.50, *p*=.003
Letter-sound knowledge (max. 32)	23.14 (7.22)	5-31	29.66 (2.68)	22-32	U=539.00, *p*<.001
Alliteration matching (max. 10)	6.25 (2.34)	1-10	8.97 (1.76)	4-10	U=496.50, *p*<.001
Nonword repetition (max. 24)	10.14 (5.45)	4-24	19.97 (3.24)	11-24	U=552.00, *p*<.001
Digit recall (max. 54)	13.67 (5.96)	6-27	26.34 (4.11)	19-37	t(33.31)=-8.41, *p*<.001
Block recall (max. 54)	14.67 (6.95)	5-26	19.07 (3.95)	11-27	t(29.28)=-2.61, *p*=.014
BPVS	50.43 (15.63)	30-87	68.86 (14.73)	40-92	t(49)=-4.50, *p*<.001
Picture naming (max.30)	19.00 (5.59)	7-27	23.62 (3.74)	15-29	t(32.27)=-3.52, *p*=.001
Sentence structure (age-equivalent score)	3;08 (1;02)	2;11-7;03	6;02 (1;09)	4;01-9;00	U=591.50, *p*<.001
MLU	4.09 (1.67)	1.40-6.67	6.19 (1.80)	2.17-10.70	t(44)=-3.91, *p*<.001
Narrative content (max. 30)	5.71 (3.39)	1-12	8.34 (2.77)	3-14	t(48)=-3.02, *p*=.004

distributions as they may affect results, for example distributions which are not normal may affect the strength of correlations, and this will be commented on where appropriate.

2.3.1.1. Group differences at Time 1.

The groups were explicitly matched on the single word reading task at Time 1, and it was expected that they would also score similarly on the early word reading task and passage reading accuracy, but that the typically developing children would score significantly better on all other tasks. Where data distributions were normal for both groups, independent samples t-tests were used and where the distribution was not normal for at least one group, a Mann-Whitney U test was conducted. The results of these analyses are shown in Table 3.

As predicted, there were no significant differences between the two groups on the three measures of reading accuracy but there were significant differences on all other tasks due to higher scores in the typically developing group. This is consistent with previous research and highlights reading accuracy as an island of ability in children with Down syndrome.

2.3.2. Descriptive Statistics at Time 2

The mean scores and standard deviations for the tasks administered at Time 2 are shown in Table 4. At Time 2, phoneme deletion was administered to all individuals who scored above chance on the alliteration matching task. This was the case for six children with Down syndrome and 28 typically developing children at Time 2. The results are not reported for children with Down syndrome due to the low numbers of children who completed this task. The results for the typically developing group are reported, as their performance on the alliteration matching task was at ceiling. Following the procedure from Time 1, MLU was not calculated for two children with Down syndrome and one typically developing child as their total number of utterances was below four.

The distributions were examined for normality as at Time 1. For the matrices task, the scores from the children with Down syndrome were clustered towards the lower end of the distribution resulting in a positive skew. Over half of the children in each group were at ceiling on the early word reading task. For both passage reading accuracy and comprehension, there was a negative skew in the scores of the typically developing children. As at Time 1, children

Table 4.

Scores of the children with Down syndrome and typically developing children on all Time 2 measures (Study 1)

	Scores of the children with Down syndrome		Scores of the typically developing children		
	Mean (standard deviation)	Range	Mean (standard deviation)	Range	Between-group differences
Matrices (age-equivalent score)	4;08 (1;02)	4;00-8;06	8;06 (2;02)	6;01-14;06	U=606.50, *p*<.001
Early word reading (max.30)	22.48 (9.30)	3-30	26.90 (5.59)	7-30	U=418.00, *p*=.047
Single word reading (max.60)	20.33 (13.45)	0-40	27.97 (13.09)	2-46	t(48)=-2.01, *p*=.050
Passage reading accuracy	32.24 (14.45)	4-52	43.70 (10.27)	15-56	U=461.00, *p*=.002
Passage reading comprehension	23.33 (14.58)	0-48	41.76 (13.15)	0-58	U=507.50, *p*<.001
Nonword reading (max. 25)	4.52 (5.04)	0-14	14.31 (7.63)	0-25	U=518.50, *p*<.001
Letter-sound knowledge (max. 32)	25.76 (5.28)	17-32	30.90 (1.32)	27-32	U=511.50, *p*<.001
Alliteration matching (max. 10)	5.81 (2.23)	1-9	9.41 (1.05)	5-10	U=607.50, *p*<.001
Phoneme deletion (max. 24)	-	-	14.11 (5.94)	0-23	-
Nonword repetition (max. 30)	10.71 (5.52)	1-20	21.90 (3.76)	8-24	U=573.00, *p*<.001
Digit recall (max. 54)	14.95 (3.83)	10-26	27.03 (4.13)	19-37	U=594.00, *p*<.001
Block recall (max. 54)	15.67 (6.50)	6-30	21.31 (3.50)	12-27	t(28.37)=-3.62, *p*=.001
BPVS	52.95 (16.72)	32-94	76.24 (14.62)	47-99	t(49)=-5.47, *p*<.001
Picture naming (max.37)	21.52 (4.91)	15-29	25.59 (3.30)	19-35	t(48)=4.687, *p*<.001
Sentence structure (age-equivalent score)	3;11 (1;05)	2;11-9;00	7;01 (1;07)	4;01-9;00	U=598.50, *p*<.001
MLU	3.91 (2.20)	1.00-8.25	6.34 (1.00)	4.44-8.80	t(23.11)=-4.52, *p*<.001
Narrative (max. 30)	6.71 (5.16)	0-19	9.35 (3.04)	3-15	t(29.96)=-2.09, *p*=.045

with Down syndrome found the nonword reading task difficult and half were at floor. For digit recall, there was a bimodal distribution for the children with Down syndrome. On both the letter-sound knowledge and alliteration task there was a ceiling effect for the typically developing group. There was a clustering of scores towards the upper end of the distribution on the nonword repetition task for the typically developing group. The children with Down syndrome generally scored quite poorly on the sentence structure task with little variation. Again, this is important to bear in mind when interpreting correlations and will be highlighted where appropriate.

2.3.2.1. Group differences at Time 2.

Independent samples t-tests and Mann-Whitney U tests were used to test between-group differences, and the results are shown in Table 4. The typically developing group performed significantly better on all tasks, with the exception of single word reading although this was marginally significant in the same direction. Therefore the groups were no longer at the same reading accuracy level. As expected the typically developing children appear to be making more progress than the children with Down syndrome.

2.3.3. Descriptive Statistics at Time 3

The scores on all tasks at Time 3 for both groups are shown in Table 5. MLU was not reported for two children with Down syndrome as they produced less than four utterances. One child with Down syndrome did not complete the narrative task and therefore MLU along with a narrative content score could not be computed. Alliteration matching and BPVS scores were not available for three children and one child with Down syndrome, respectively, because of refusal to complete the task.

The distributions were examined for normality as previously. For the passage reading accuracy, there were some outliers in the group of children with Down syndrome towards the lower end of the distribution causing a positive skew. On the nonword reading task, there was clustering of scores towards the higher end of the distribution in the typically developing group and clustering towards the lower end for the children with Down syndrome. As would be expected

Table 5.

Scores of the children with Down syndrome and typically developing children on all Time 3 measures (Study 1)

	Scores of the children with Down syndrome		Scores of the typically developing children		
	Mean score (standard deviation)	Range	Mean score (standard deviation)	Range	Between-group differences
Single word reading (max.60)	23.19 (14.39)	1-45	33.86 (10.60)	13-48	t(34.96)=-2.88, *p*=.007
Passage reading accuracy	33.38 (15.14)	3-54	50.00 (8.46)	32-64	U=518.00, *p*<.001
Passage reading comprehension	26.24 (14.67)	0-59	49.93 (9.68)	28-68	t(32.30)=-6.45, *p*<.001
Nonword reading (max. 25)	6.00 (6.65)	0-19	16.90 (7.27)	5-25	U=531.00, *p*<.001
Alliteration matching (max. 10)	6.78 (2.26)	3-10	9.48 (1.18)	4-10	U=466.50, *p*<.001
Phoneme deletion (max. 24)	-	-	16.52 (5.77)	4-24	-
BPVS	56.90 (19.07)	31-98	85.62 (12.13)	60-105	t(48)=-6.71, *p*<.001
Picture naming (max.37)	22.57 (6.34)	11-31	28.90 (4.78)	20-36	t(32.60)=-3.89, *p*<.001
MLU	4.40 (2.19)	1.00-7.89	7.07 (1.84)	1.60-10.40	t(45)=-4.51, *p*<.001
Narrative (max.30)	6.95 (3.63)	2-15	10.62 (3.52)	4-19	t(47)=-3.54, *p*<.001

from previous time-points, there was a ceiling effect on the alliteration matching task in the typically developing group.

2.3.3.1. Group differences at Time 3.

The results from the between-group analyses, either independent samples t-tests or Mann-Whitney U tests, are shown in Table 5. The typically developing children continued to make greater progress than the children with Down syndrome and therefore scored significantly higher on all measures.

2.3.4. Concurrent relationships at Time 1.

At Time 1, the groups performed comparably on all three reading accuracy measures, and as can be seen from Table 6, these measures were highly correlated with each other in both groups. Therefore a reading accuracy composite score was formed by averaging z-scores from these three measures. All further analyses were performed using the reading accuracy composite score. A vocabulary composite was also formed as the two measures of vocabulary correlated highly. This is presented in correlations along with the individual tasks as it will be used when presenting path models, therefore it is important to know the correlations involving this variable.

Table 6.

Time 1 correlations between the individual reading measures for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Early word reading	Single word reading	Passage reading accuracy
Early word reading	-	.86**	.86**
Single word reading	.91**	-	.84**
Passage reading accuracy	.93**	.91**	-

*p<0.05 **p<0.01

Table 7 shows the Pearson correlation coefficients for both groups. The matrices task was moderately to strongly correlated with other cognitive variables in both groups, as was chronological age for the typically developing children. Therefore correlations were also conducted partialling for age and nonverbal ability (Table 8). The following descriptions will be based on simple correlations and notable differences in the partial correlations will be highlighted. The significance or otherwise of the correlations will be commented on. The sample sizes are unequal and therefore the power to detect a significant result is different in the two groups therefore the strength of the relationships will also be discussed. The strength of the correlations were tested for between-group differences for these and all subsequent sets of correlations, and any significant differences are noted in the corresponding tables.

The two groups were matched on reading accuracy at Time 1 only and were therefore also reading similar passages from which their reading comprehension scores were calculated. At the subsequent time-points, and most importantly at Time 3 which will be used for the longitudinal analyses, the typically developing children were reading at a higher level. Therefore the concurrent relationships at Time 1 are important as these can reveal differences between children with Down syndrome and typically developing children which cannot be due to their stage of reading accuracy development. Therefore for both reading accuracy and reading comprehension, path models based on simultaneous regression were conducted. To control for age and nonverbal ability, raw scores were residualised for age and matrices and these standardised residuals were used in the path models. To test whether any of the pathways were significantly different in the two groups, correlations partialling for the other variable in the model were computed and tested for between-group differences. Where there were significant differences, this is reported.

2.3.4.1. Hearing.

The correlations of the tasks with hearing threshold levels were examined for the children with Down syndrome. The only significant correlation was between hearing threshold at 1000Hz and letter-sound knowledge ($r=-.65$), and therefore hearing threshold will not be included in any further analyses.

Table 7.

Time 1 simple correlations between all measures for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Age	Matrices	Reading accuracy	Nonword reading	Reading comprehension	Letter-sound knowledge	Alliteration matching	Nonword repetition	Digit recall	Block recall	Picture naming	BPVS	Vocabulary composite	Sentence structure	MLU	Narrative content
Age	-	.24	.30†	.03†	.22	-.20	.18	-.06	-.09	.20	.32	.39	.38	.30	-.31†	.31
Matrices	.62**	-	.43[a]	.60**	.47*	-.03†	.36	.54*	.59**	.50*	.59**	.76**	.71**	.84**†	.57*	.31
Reading accuracy	.87**†	.71**	-	.65**†	.83**	.44*	.38	.53*	.43[b]	.32	.75**	.54*	.70**	.46*	.49*	.26
Nonword reading	.78**†	.67**	.91**†	-	.55**	.36	.40	.49*	.60**	.33	.67**	.49*	.63**	.71**	.59*	.37
Reading comprehension	.57**	.59**	.76**	.71**	-	.31	.19	.64**	.43[b]	.23	.71**	.55**	.69**	.56**	.54*	.33
Letter-sound knowledge	.47*	.55**†	.64**	.62**	.50**	-	.16†	.16	.15	-.10†	.28	.14†	.23	.15	.14	.20
Alliteration matching	.46*	.43*	.63**	.53**	.59**	.75**†	-	.33	.56*	.74**†	.53*	.60**	.60**	.42	.60*	.40
Nonword repetition	.42*	.45*	.61**	.56**	.78**	.46*	.57**	-	.81**†	.50*	.68**	.66**	.72**	.60**	.75**	.49*
Digit recall	.23	.54**	.35	.31	.18	.50**	.33	.25†	-	.61**	.68**	.61**	.69**	.67**	.91**†	.36
Block recall	.36	.39*	.45*	.35	.49**	.52**†	.27†	.45*	.25	-	.64**	.64**	.68**	.50*	.53*	.37
Picture naming	.43*	.51**	.58**	.52**	.66**	.49**	.56**	.69**	.53**	.26	-	.76**	.95**	.58**	.69**	.61**
BPVS	.53**	.51**	.67**	.66**	.68**	.66**†	.70**	.69**	.48**	.39*	.77**	-	.93**	.73**	.51*	.58**
Vocabulary composite	.51**	.54**	.67**	.63**	.68**	.61**	.68**	.73**	.54**	.34	.93**	.95**	-	.69**	.65**	.64**
Sentence structure	.63**	.46*†	.72**	.68**	.64**	.45*	.38*	.57**	.44*	.56**	.61**	.69**	.70**	-	.57*	.44*
MLU	.57**†	.39*	.55**	.53**	.37*	.34	.20	.40*	.28†	.18	.48**	.51**	.53**	.57**	-	.80**
Narrative content	.33	.45*	.53**	.55**	.47*	.44*	.32	.56**	.46*	.18	.68**	.60**	.68**	.55**	.63**	-

Note. [a]p=.051 [b]p=.052

†significant difference between the correlations in the two groups

*p<0.05 **p<0.01

Table 8.

Time 1 partial correlations between all measures controlling for age and matrices for the children with Down syndrome (above the diagonal) and the typically developing group (below the diagonal) (Study 1)

	Word reading	Nonword reading	Reading comprehension	Letter-sound knowledge	Alliteration matching	Nonword repetition	Digit recall	Block recall	Picture naming	BPVS	Vocabulary composite	Sentence structure	MLU	Narrative content
Word reading	-	.59**	.78**	.57*	.25	.47*	.33	.11	.67**	.31	.60**	.17	.57*	.10
Nonword reading	.68**	-	.41	.46*	.27	.22	.36	.05	.54*	.11	.42	.52*	.36	.30
Reading comprehension	.59**	.44*	-	.40	.01	.57*	.26	-.02	.60**	.32	.55*	.33	.54*	.20
Letter-sound knowledge	.46*	.37[a]	.34	-	.20	.17	.15	-.08	.43	.34	.44[b]	.36	.09	.29
Alliteration matching	.50**	.26	.43*	.66**	-	.21	.51*	.68** †	.41	.54*	.52*	.22	.70** †	.31
Nonword repetition	.48*	.35	.64**	.26	.44*	-	.71** †	.35	.62**	.59**	.68**	.38	.64*	.49*
Digit recall	.11	.03	-.26	.33	.19	.04 †	-	.51*	.62**	.45[c]	.62**	.47*	.88** †	.34
Block recall	.22	.04	.31	.38[d]	.08 †	.31	.09	-	.49*	.45[e]	.53*	.15	.47	.25
Picture naming	.37	.22	.40*	.26	.42*	.58**	.39*	.05	-	.57*	.94**	.17	.82** †	.53*
BPVS	.45*	.42*	.50**	.50**	.59**	.58**	.36	.20	.68**	-	.82**	.21	.42	.53*
Vocabulary composite	.45*	.35	.50**	.42*	.56**	.63**	.41*	.14	.91**	.92**	-	.21	.75**	.59**
Sentence structure	.44*	.38[e]	.36	.20	.10	.42*	.39*	.45*	.47*	.54**	.56**	-	.40	.32
MLU	.13	.14	.13	.09	-.11 †	.21	.19 †	-.05	.32 †	.30	.33	.32	-	.03 †
Narrative content	.44*	.42*	.34	.25	.14	.44*	.30	.00	.58**	.49*	.58**	.45*	.59** †	-

Note. [a]p=.054 [b]p=.058 [c]p=.056 [d]p=.053 [e]p=.051

†significant difference between the correlations in the two groups

*p<0.05 **p<0.01

2.3.4.2. Reading accuracy.

2.3.4.2.1. Concurrent correlations.

At Time 1, letter-sound knowledge and alliteration matching were moderately and significantly related to reading accuracy in the typically developing group. For the children with Down syndrome, letter-sound knowledge was significantly correlated with reading accuracy but alliteration matching was not. Notably, the correlation between alliteration matching and letter-sound knowledge was significantly stronger for the typically developing children than for the children with Down syndrome. Reading accuracy and nonword reading were significantly correlated in both groups. This relationship in the typically developing group was significantly stronger than for the children with Down syndrome in the simple but not partial correlations, indicating that this is not a robust finding and some of the shared variance between these two measures in the typically developing children was due to general ability.

In the simple correlations, reading accuracy was correlated at a moderate to strong level with all of the oral language measures in both groups, with the exception of narrative content in the group of children with Down syndrome. In the partial correlations, nonword repetition and the vocabulary composite were significantly correlated with reading accuracy in both groups, although for the individual measures of vocabulary the correlations were significant for the receptive task in the typically developing group and the expressive task for the group of children with Down syndrome. Sentence structure and narrative content were significantly related to reading accuracy in the typically developing group only and MLU was significantly correlated with reading accuracy in the group of children with Down syndrome only.

In early typical development, MLU and vocabulary are closely linked and expressive vocabulary has been found to strongly predict MLU for pre-school children (Dethorne et al., 2005; Devescovi et al., 2005). As the children with Down syndrome who participated in the present study are of a similar mental age to the groups of typically developing children in these studies, it may be that MLU does not relate to reading accuracy independently of picture naming, a measure of expressive vocabulary. Indeed the correlation between the two measures was strong. Therefore partial correlations were computed controlling for picture naming and the relationship between MLU and reading accuracy for the children with Down syndrome was now low and not significant (r=-.06). However if MLU was controlled for in a partial correlation, the relationship between picture naming and reading accuracy remained strong and significant (r=.66). Therefore, MLU seems to relate to reading accuracy in children with Down syndrome via picture naming, not independently.

In the simple correlations the memory measures were similarly correlated with reading accuracy in both groups, but only the correlation between block recall and reading accuracy in the typically developing group was significant. However in the partial correlations, this was no longer significant.

2.3.4.2.2. Path models.

From previous research and the pattern in the concurrent correlations, phoneme awareness and oral language were selected to be of interest in predicting reading accuracy. Phoneme awareness was assessed using alliteration matching at Time 1 and the vocabulary composite was chosen to reflect oral language as this correlated with reading accuracy in both groups. Path models predicting reading accuracy (Read1) from alliteration matching (PA1) and vocabulary (Vocab1) are shown in Figure 3. Dashed lines represent beta weights which were not significant and solid lines represent beta weights which were significant. In the typically developing group, neither phoneme awareness nor vocabulary were significant predictors of reading accuracy, although p=.078 for the phoneme awareness pathway. For the children with Down syndrome, vocabulary was a significant predictor of reading accuracy, whereas phoneme awareness was not.

2.3.4.2.3. Summary: Reading accuracy at Time 1.

The finding that phoneme awareness is significantly correlated with reading accuracy in typically developing children but not children with Down syndrome was replicated here. In the path model, phoneme awareness did not add unique variance to reading accuracy in either group, although there was a trend for this to be significant in the typically developing children. In the correlations, letter-sound knowledge had a moderate relationship with word and nonword reading in both groups. Furthermore word and nonword reading were significantly correlated with each other in both groups.

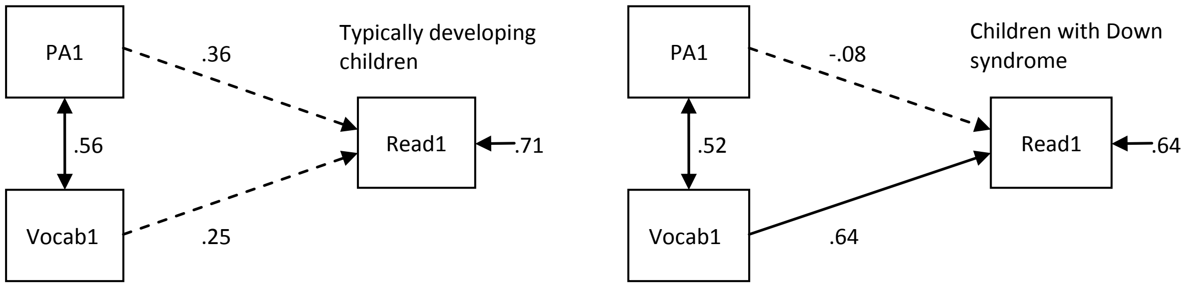


Figure 3. Path models using simultaneous regression and residuals standardised for age and nonverbal ability to predict Time 1 reading accuracy with Time 1 phoneme awareness and vocabulary as predictors (Study 1)

The vocabulary composite and nonword repetition were correlated with reading accuracy in both groups, as was narrative content in the typically developing group and MLU in the children with Down syndrome. Within vocabulary, BPVS was correlated with reading accuracy in the typically developing group and picture naming was related to reading accuracy in the children with Down syndrome. In the path models, vocabulary did not add unique variance to reading accuracy in the typically developing group but this pathway was significant for the children with Down syndrome.

It should be noted that there were no significant differences between the two groups regarding the relationship between reading accuracy and phoneme awareness, and reading accuracy and oral language in either the correlations or path models. The small sample size in the present study may result in a lack of power to detect significant differences between the two groups.

2.3.4.3. Reading comprehension.

2.3.4.3.1. Concurrent correlations.

Reading comprehension was significantly correlated with reading accuracy in both groups in the simple and partial correlations. The simple correlations with the language measures were moderate to high in both groups, and significant with the exception of narrative content in the children with Down syndrome. In the partial correlations, nonword repetition and the vocabulary composite remained significantly correlated with reading comprehension in both groups. Considering the two components of vocabulary, both tasks were significantly correlated with reading comprehension in the typically developing group, whereas only the relationship with picture naming was significant for the children with Down syndrome. Neither sentence structure nor narrative content had a significant relationship with reading comprehension in either group and MLU was significantly correlated in the group of children with Down syndrome only.

The pattern of relationship with memory differs in the two groups in the simple correlations, with digit recall being marginally significant in the group of children with Down syndrome and block recall being significant in the typically developing group. In the partial correlations however, none of these correlations were significant.

2.3.4.3.2. Path models.

The simple view of reading was used as a framework for reading comprehension path models, therefore reading accuracy (Read1) and the vocabulary composite at Time 1 (Vocab1) were entered as predictors for reading comprehension (Comp1). These path models are shown in Figure 4. For both groups of children, the pathway from reading accuracy to reading comprehension was significant, but vocabulary did not add unique variance.

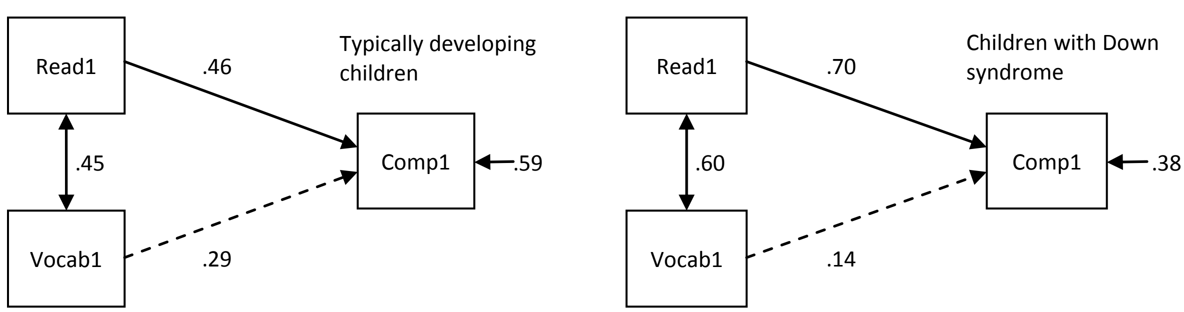


Figure 4. Path models using simultaneous regression and residuals standardised for age and nonverbal ability to predict Time 1 reading comprehension with Time 1 reading accuracy and vocabulary as predictors (Study 1)

2.3.4.3.3. Summary: Reading comprehension at Time 1.

Reading comprehension was similarly correlated with word and nonword reading in the two groups. Concerning the language measures, the vocabulary composite was related to reading comprehension in both groups, along with MLU for the children with Down syndrome. Within the vocabulary composite, reading comprehension was correlated with both tasks for the typically developing children and only picture naming for the children with Down syndrome. Digit recall was not related to reading comprehension in either group. The path models indicate that when the relative contribution of reading accuracy and oral language is considered at this early stage in literacy development, only reading accuracy has a unique influence on reading comprehension.

2.3.5. Concurrent Relationships at Time 2

As at Time 1, the reading accuracy and vocabulary composites were formed. As can be seen in Table 9 the three reading accuracy measures were highly inter-correlated, and thus all further analyses were performed using the reading accuracy composite. Simple correlations are shown in Table 10 and correlations partialling for age and matrices are shown in Table 11, and the strength of the correlations tested for between-group differences. The groups are no longer at the same reading accuracy level; therefore only correlations will be described highlighting key similarities and differences from Time 1.

Table 9.

Time 2 correlations between the individual reading measures for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Early word reading	Single word reading	Passage accuracy
Early word reading	-	.87**	.92**
Single word reading	.78**	-	.87**
Passage accuracy	.81**	.88**	-

2.3.5.1. Reading accuracy.

The most notable difference from Time 1 is the weak relationship between reading accuracy and both letter-sound knowledge and alliteration matching in the typically developing group. As evident in the descriptive scores for Time 2, there are ceiling effects for both these tasks for the typically developing children. Phoneme deletion was a more sensitive measure of phonological awareness for this group of children, and indeed the scores on this task were highly correlated with the reading accuracy composite. Due to the weak correlation between letter-sound knowledge and reading accuracy in the typically developing group, this relationship was significantly stronger for the children with Down syndrome. Nonword reading remained strongly correlated with word reading in both groups.

The simple correlations with oral language were all significant, with the exception of nonword repetition and MLU in the typically developing group. The partial correlations were similar to the simple correlations. For both groups, the vocabulary composite (along with each individual task) was significantly related to reading accuracy, but nonword repetition and sentence structure were not. For the children with Down syndrome only, MLU and narrative content were also significantly related to reading accuracy.

Digit recall was significantly correlated with reading accuracy in both groups, and block recall was not. In the partial correlations, the relationship between digit recall and reading accuracy remained significant for the children with Down syndrome.

2.3.5.2. Reading comprehension.

Reading comprehension was significantly correlated with reading accuracy and nonword reading in both groups, and in both the simple and partial correlations. In the simple correlations, all of the oral language measures were correlated at moderate to high levels with reading comprehension for the children with Down syndrome. In the typically developing

Table 10.
Time 2 simple correlations between all measures for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Age	Matrices	Reading accuracy	Nonword reading	Reading comprehension	Letter-sound knowledge	Alliteration matching	Phoneme deletion	Nonword repetition	Digit recall	Block recall	Picture naming	BPVS	Vocabulary composite	Sentence structure	MLU	Narrative content
Age	-	.31	.15	.01 †	.19	-.26	.13	-	.00	.12	.18	.25	.20	.23	.27	.07	.26
Matrices	.48**	-	.41	.45*	.51*	.14	.43*	-	.47*	.37	.62**	.60**	.78**	.71**	.69**	.43	.64**
Reading accuracy	.63**	.51**	-	.71**	.77**	.64** †	.30	-	.51*	.58**	.06	.78**	.68**	.75**	.44*	.66**	.57**
Nonword reading	.60** †	.52**	.86**	-	.76**	.60**	.58**	-	.56**	.58**	.15	.61**	.63**	.64**	.59**	.52*	.46*
Reading comprehension	.51**	.48**	.79**	.64**	-	.46* †	.61**	-	.61**	.72**	.32	.77**	.75**	.78**	.54*	.70**	.66**
Letter-sound knowledge	-.21	.19	.10 †	.11	-.14 †	-	.05	-	.16	.31	-.18	.50*	.42	.47*	.34	.62**	.33
Alliteration matching	.02	.19	.32	.33	.48**	-.02	-	-	.53*	.57**	.55**	.44**	.49*	.48*	.35	.46*	.56**
Phoneme deletion	.49**	.38*	.77**	.79**	.80**	-.07	.52**	-	-	-	-	-	-	-	-	-	-
Nonword repetition	.18	.07	.29	.29	.43*	-.15	.01	.49**	-	.73** †	.50* †	.58**	.57**	.58**	.59**	.38	.50*
Digit recall	.30	.45*	.48**	.62**	.33	.15	.24	.29	.20 †	-	.55**	.67** †	.64**	.68** †	.63**	.70** †	.64**
Block recall	.26	.36[a]	.31	.32	.29	.11	.24	.15	-.19 †	.33	-	.44*	.56**	.52*	.38	.34	.44*
Picture naming	.45*	.35	.69**	.53**	.77**	-.04	.41*	.59**	.35	.19 †	.15	-	.88**	.97**	.60**	.84** †	.72** †
BPVS	.61**	.52**	.77**	.68**	.74**	-.08	.30	.76**	.36[b]	.37*	.13	.67**	-	.97**	.76**	.79** †	.76** †
Vocabulary composite	.58**	.47**	.80**	.66**	.83**	-.06	.39*	.73**	.37[b]	.30 †	.15	.92**	.91**	-	.70**	.84** †	.76** †
Sentence structure	.55**	.42*	.60**	.53**	.60**	-.01	.37*	.50**	.09	.25	.53**	.53**	.60**	.61**	-	.57*	.66**
MLU	.27	.25	.35	.33	.32	.24	.36	.27	-.01	.04 †	.16	.23 †	.16 †	.21 †	.24	-	.74**
Narrative content	.15	.36[c]	.38*	.40*	.36[d]	.21	.39*	.24	.08	.33	.51**	.21 †	.11 †	.17 †	.21	.40*	-

Note. [a]p=.053 [b]p=.052 [c]p=.056 [d]p=.055
†significant difference between the correlations in the two groups
*p<0.05 **p<0.01

Table 11.
Time 2 partial correlations controlling for age and matrices for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Reading accuracy	Nonword reading	Reading comprehension	Letter-sound knowledge	Alliteration matching	Phoneme deletion	Nonword repetition	Digit recall	Block recall	Picture naming	BPVS	Vocabulary composite	Sentence structure	MLU	Narrative content
Reading accuracy	-	.66**	.72**	.69**†	.15	-	.41	.50*	-.26	.73**	.64**	.72**	.23	.59*	.44[a]
Nonword reading	.76**	-	.70**	.60**	.48*	-	.42	.51*	-.19	.50*	.50*	.52*	.46*	.39	.27
Reading comprehension	.67**	.42*	-	.50*†	.50*	-	.51*	.66**†	.00	.67**	.66**	.69**	.30	.63**	.51*
Letter-sound knowledge	.22†	.22	-.16†	-	-.01	-	.06	.30	-.36	.58**†	.50*†	.57*	.39	.64**	.37
Alliteration matching	.35	.36	.53**	-.10	-	-	.41	.49*	.40	.26	.27	.27	.08	.34	.41
Phoneme deletion	.67**	.70**	.73**	-.03	.57**	-	-	-	-	-	-	-	-	-	-
Nonword repetition	.25	.25	.43**	-.12	.01	.48*	-	.68**†	.30	.45[b]	.36	.43	.44	.21	.31
Digit recall	.32	.52**	.11†	.12	.19	.11	.18†	-	.44	.61**†	.61**	.64**	.55*	.64**†	.56*
Block recall	.12	.13	.12	.09	.19	-.03	-.26	.19	-	.12	.17	.15	-.08	.09	.07
Picture naming	.56**	.33	.69**	.01†	.42*	.45*	.32	-.01†	-.02	-	.83**	.97**	.31	.82**†	.53*
BPVS	.59**	.43*	.59**	-.06†	.32	.65*	.35	.14	-.14	.54**	-	.95**	.50*	.80**†	.57*†
Vocabulary composite	.65**	.42*	.74**	-.02	.43*	.61**	.38[c]	06	-.08	.91**	.85**	-	.41	.84**	.57*
Sentence structure	.35	.24	.40*	.07	.41*	.28	-.01	.03	.44*	.35	.36	.40*	-	.43	.39
MLU	.21	.18	.18	.29	.35	.14	-.06	-.10†	.06	.10†	-.06†	.03	.09	-	.68**
Narrative content	.31	.31	.26	.15	.35	.14	.07	.20	.44*	.11	-.10†	.02	.10	.36	-

Note. [a]p=.058 [b]p=.055 [c]p=.052
†significant difference between the correlations in the two groups
*p<0.05 **p<0.01

group, all of the measures except MLU and narrative content were significantly correlated with reading comprehension. In the partial correlations, vocabulary and nonword repetition were significantly related to reading comprehension in both groups. Reading comprehension was also significantly correlated with MLU and narrative content in the children with Down syndrome and sentence structure in the typically developing group.

Of the memory measures, digit recall was highly correlated to reading comprehension in the children with Down syndrome. This relationship was significantly stronger than in the typically developing group in partial correlations.

2.3.5.3. Summary: Concurrent relationships at Time 2.

At Time 2, the correlations with reading accuracy were very similar to Time 1 for the children with Down syndrome. This is likely to be because they made little progress on the reported tasks compared to the typically developing children. The main difference from Time 1 for the children with Down syndrome is the significant relationship with digit recall; additionally the trend for BPVS to be related to reading accuracy is now significant. Reading accuracy remained significantly correlated with nonword reading, letter-sound knowledge, picture naming and MLU. In comparison for the typically developing children, due to ceiling effects alliteration matching and letter-sound knowledge were no longer significant correlated with reading accuracy. In comparison, phoneme deletion was related to reading accuracy and within the language measures, vocabulary appeared to be most important. The only significant difference between the two groups was regarding letter-sound knowledge which was more strongly correlated with reading accuracy in the group of children with Down syndrome.

Reading comprehension was similarly related to reading accuracy in both groups. The correlations with the language measures were moderate to high and similar to Time 1. A notable difference was the relationship with digit recall for the children with Down syndrome, which was significantly stronger than in the typically developing group.

2.3.6. Concurrent Relationships at Time 3

At Time 3 two reading accuracy measures (single word reading and passage reading accuracy) were administered and the correlations between these were very high for the children with Down syndrome and the typically developing group (r=.88 for both groups) so a reading accuracy composite was calculated. A vocabulary composite was also formed. The simple correlations for Time 3 are shown in Table 12 and correlations partialling for age (matrices was not administered at Time 3) are shown in Table 13.

2.3.6.1. Reading accuracy.

The correlations between reading accuracy and both nonword reading and phonological awareness were similar to Time 2. Nonword reading was significantly correlated with reading in both groups. Phoneme deletion was significantly related to reading in the typically developing group and alliteration matching was marginally significant in the children with Down syndrome in the simple correlations although this reduced to a weak relationship in the partial correlations. Considering the oral language measures, the vocabulary tasks were significantly correlated with reading accuracy in both groups, with the addition of narrative content for the typically developing children in the simple correlations only.

2.3.6.2. Reading comprehension.

Reading comprehension was similarly related to word and nonword reading in both groups, mirroring the pattern seen at previous time-points. In the simple correlations, all the oral language measures were significantly correlated with reading comprehension in the typically developing group; in the partial correlations the correlation with MLU was no longer significant. For the children with Down syndrome, only the vocabulary measures were significantly related to reading comprehension, and the strength of these correlations was high.

2.3.6.3. Summary: Concurrent relationships at Time 3.

The pattern of correlations for the reading accuracy composite in both groups was clear. Phonological awareness was moderately correlated in the children with Down syndrome, although this was not significant. However in the typically developing group this relationship was stronger and significant, although this was not significantly different than for the children with Down syndrome. The two vocabulary measures were correlated significantly with reading accuracy at a similar strength in both groups.

Table 12.

Time 3 simple correlations between all measures for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Age	Reading accuracy	Nonword reading	Reading comprehension	Alliteration matching	Phoneme deletion	Picture naming	BPVS	Vocabulary composite	MLU	Narrative content
Age	-	.12[†]	.12	-.01	.31	-	.14	.20	.11	-.06	.36
Reading accuracy	.64**[†]	-	.77**	.66**	.46[a]	-	.70**	.62**	.69**	.43	.01
Nonword reading	.44*	.79**	-	.60**	.28	-	.75**	.55*	.68**	.43	.18
Reading comprehension	.36[b]	.68**	.60**	-	.56*	-	.76**	.79**	.81**	.43	-.02
Alliteration matching	-.12	.17	.19	.34	-	-	.64**	.68**[†]	.71**	.37	.11
Phoneme deletion	.44*	.78**	.70**	.66**	.06	-	-	-	-	-	-
Picture naming	.46*	.73**	.53**	.66**	.24	.67**	-	.79**	.96**	.53*	.13
BPVS	.55**	.70**	.45*	.61**	-.01[†]	.63**	.85**	-	.94**	.53*	.26
Vocabulary composite	.52**	.75**	.52**	.67**	.14	.68**	.97**	.95**	-	.58*	.15
MLU	.35	.34	.12	.42*	-.03	.28	.17	.31	.24	-	.55*
Narrative content	.43*	.53**	.54**	.48**	-.04	.50**	.32	.44*	.39*	.47*	-

Note. [a]p=.056 [b]p=.055

[†]significant difference between the correlations in the two groups

*p<0.05 **p<0.01

Table 13.

Time 3 partial correlations controlling for age between all measures for the children with Down syndrome (above the diagonal) and the typically developing children (below the diagonal) (Study 1)

	Reading accuracy	Nonword reading	Reading comprehension	Alliteration matching	Phoneme deletion	Picture naming	BPVS	Vocabulary composite	MLU	Narrative content
Reading accuracy	-	.76**	.67**	.45	-	.69**	.61**	.68**	.44	.06
Nonword reading	.74**	-	.60**	.26	-	.75**	.54*	.68**	.44	.15
Reading comprehension	.62**	.52**	-	.59*	-	.77**	.81**	.82**	.43	-.02
Alliteration matching	.33	.28	.42*	-	-	.63**	.66**†	.71**	.41	.00
Phoneme deletion	.73**	.63**	.59**	.12	-	-	-	-	-	-
Picture naming	.64**	.41*	.60**	.34	.58**	-	.79**	.95**	.55*†	.08
BPVS	.54**	.27	.53**	.07†	.51**	.80**	-	.95**	.56*	.20
Vocabulary composite	.63**	.37[b]	.60	.24	.58**	0.96**	.93**	-	.59*	.12
MLU	.16	-.04	.33	.01	.15	.01†	.15	.07	-	.61**
Narrative content	.37[a]	.44*	.38*	.02	.39*	.15	.26	.21	.38*	-

Note. [a]p=.051 [b]p=.050

†significant difference between the correlations in the two groups

*p<0.05 **p<0.01

Reading comprehension was significantly related to reading accuracy and phonological awareness in both groups. The vocabulary tasks were strongly correlated with reading comprehension for the children with Down syndrome and significantly but at a slightly lower level for the typically developing children.

2.3.7. Longitudinal Relationships

The longitudinal analyses with reading accuracy, reading comprehension and oral language as outcome measures at Time 3 will now be presented. The time period from Time 1 to Time 3 will be focussed on as this allows for maximum growth. At Time 1, the children with Down syndrome had a mean age of 11;05 and the typically developing children had a mean age of 6;02. Time 3 took place almost two years later and the children with Down syndrome had a mean age of 13;02 and the typically developing children had a mean age of 7;11. At Time 1 the groups were matched on reading accuracy, but due to greater progress by the typically developing children this was no longer the case at Times 2 and 3.

For each outcome measure, the correlations from Time 1 to Time 3 will be shown, and path models based on simultaneous regression will be conducted as for the concurrent relationships at Time 1. Raw scores residualised for age were used in the path models. Nonverbal ability could not be controlled for in the same manner as it was not assessed at Time 3. A maximum of three predictors will be entered and this should still be interpreted with caution in light of the relatively small sample size. Correlations partialling for the other variables in the model were computed and tested for between-group differences and significant differences will be highlighted.

2.3.7.1. Predicting reading accuracy.

The simple correlations between Time 3 reading accuracy and Time 1 measures are shown in Table 14. Notably the relationship between Time 1 and Time 3 reading accuracy was very strong in both groups. In general the correlations between reading accuracy and most other variables were moderate to strong in both groups.

When partial correlations controlling for Time 1 matrices and age were computed, alliteration matching was significantly related to reading accuracy in the typically developing

Table 14.
Correlations between Time 1 measures and Time 3 reading accuracy for the children with Down syndrome and the typically developing children (Study 1)

	Simple correlations		Partial correlations controlling for Time 1 age and matrices		Partial correlations controlling for Time 1 reading accuracy	
	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children
Age	.12	.64**	-	-	-.53**	-.29
Matrices	.35	.69**	-	-	-.18	.26
Reading accuracy	.94**	.83**	.96**†	.64**†	-	-
Nonword reading	.63**	.81**	.58**	.57**	.10	.25
Reading comprehension	.83**	.75**	.81**	.54**	.28	.35
Letter-sound knowledge	.55**	.65**	.62**	.42*	.46*	.28
Alliteration matching	.33	.63**	.23	.45*	-.08	.23
Nonword repetition	.55*	.60**	.48*	.41*	.18	.23
Digit recall	.51*	.45*	.43	.21	.35	.31
Block recall	.27	.42*	.12	.18	-.09	.09
Picture naming	.74*	.66**	.72**	.48*	.14	.40*
BPVS	.47*	.67**	.33	.45*	-.15	.27
Vocabulary composite	.66**	.71**	.64**	.51**	-.01	.37[b]
Sentence structure	.42	.67**	.25	.44*	-.04	.18
MLU	.60*	.44*	.65**†	.11†	.48[a]	-.04
Narrative content	.22	.54**	.12	.36	-.10	.22

Note. [a] p=.052 [b] p=.059
†significant difference between the correlations in the two groups
*p<0.05 **p<0.01

group only, although letter-sound knowledge was significantly correlated in both groups. Concerning the oral language measures, the vocabulary composite, MLU and nonword repetition were significantly correlated with reading accuracy in the children with Down syndrome, whereas in the typically developing group, sentence structure, the vocabulary composite and nonword repetition were related to reading accuracy. Considering the vocabulary measures individually, only picture naming was significantly correlated with later reading accuracy for the children with Down syndrome, and both measures were for the typically developing children. Memory was not significantly related to reading accuracy in either group. For the children with Down syndrome reading accuracy at Time 3 was correlated with Time 1 reading and MLU at a significantly higher level than in the typically developing group.

Earlier and later reading accuracy was correlated highly in both groups, especially for the children with Down syndrome. Therefore when reading accuracy at Time 1 was partialled out many of the remaining correlations reduced to a weak and no longer significant level. The exceptions were age for the children with Down syndrome, which had a negative relationship with reading accuracy and letter-sound knowledge and picture naming in the typically developing group.

Simultaneous regressions were conducted to evaluate the unique variance that Time 1 predictors accounted for in Time 3 reading accuracy scores. Reading accuracy at Time 1 was selected to control for autoregressive effects. As at Time 1, phoneme awareness as assessed with alliteration matching and oral language as measured by the vocabulary composite were chosen. Path models predicting reading accuracy at Time 3 (Read3) from Time 1 measures of reading accuracy (Read1), phoneme awareness (PA1) and vocabulary (Vocab1) are shown in Figure 5. The only significant pathway from Time 1 to Time 3 is the autoregressor in both groups; neither phonological awareness nor vocabulary added unique variance. This is unsurprising given the strong correlations between reading accuracy at the two time-points, and the relationship was significantly stronger for the children with Down syndrome compared to the typically developing children.

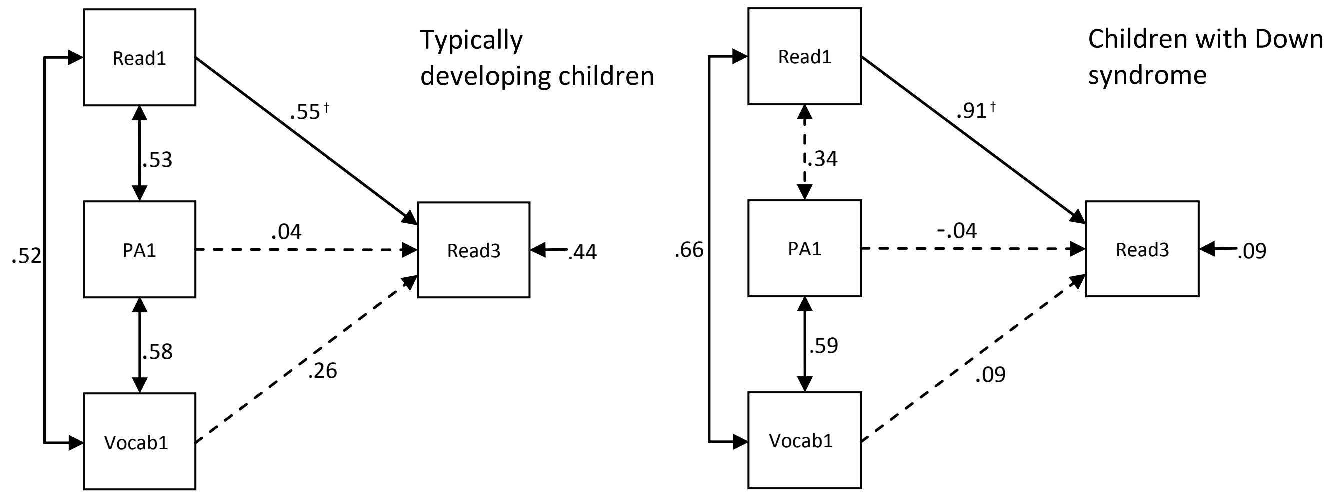


Figure 5. Path models using simultaneous regression and residuals standardised for age to predict Time 3 reading accuracy with Time 1 reading accuracy, phoneme awareness and vocabulary as predictors (Study 1)

†significant difference between the correlations in the two groups

In summary, correlations indicated that reading accuracy was significantly related to earlier phoneme awareness in the typically developing group, and measures of oral language in both groups, although the correlation with MLU was significantly higher for the children with Down syndrome. Initial levels of reading were also strongly correlated with Time 3 reading accuracy and when these were accounted for in a path model there was little variance remaining in either group and neither vocabulary nor phoneme awareness added unique variance in either group.

2.3.7.2. Predicting reading comprehension.

The correlations between Time 1 measures and Time 3 reading comprehension are shown in Table 15. In the simple correlations most relationships were significant. Word and nonword reading were similarly correlated with reading comprehension in both groups. Digit recall and MLU were both more strongly related to reading comprehension for the children

Table 15.
Correlations between Time 1 measures and Time 3 reading comprehension for the children with Down syndrome and the typically developing children (Study 1)

	Simple correlations		Partial correlations, controlling for Time 1 age and matrices		Partial correlations controlling for Time 1 reading comprehension	
	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children
Age	-.01	.35	-	-	-.21	-.10
Matrices	.51*	.52**	-	-	.30	.18
Word reading	.58**	.65**	.53*	.70**	.11	.24
Nonword reading	.59**	.61**	.39	.49**	.36	.20
Reading comprehension	.65**	.72**	.57*	.62**	-	-
Letter-sound knowledge	.21	.52**	.24	.32	.01	.18
Alliteration matching	.58**	.51**	.52*	.38*	.61**	.14
Nonword repetition	.72**	.67**	.60**	.57**	.52*	.29
Digit recall	.79**†	.40*†	.69**†	.18†	.74**	.47*
Block recall	.55*	.26	.42	.07	.54*†	-.15†
Picture naming	.67**	.63**	.59**	.50**	.38	.37[a]
BPVS	.69**	.69**	.65**	.60**	.53*	.40*
Vocabulary composite	.72**	.71**	.68**	.61**	.50*	.43*
Sentence structure	.61**	.56**	.45[b]	.46*	.40	.22
MLU	.84**†	.41*†	.82**†	.27†	.74**†	.17†
Narrative content	.26	.68**	.18	.58**	.07	.52**

Note. [a] p=.055 [b] p=.056
†significant difference between the correlations in the two groups
*p<0.05 **p<0.01

with Down syndrome than the typically developing group. The correlation with vocabulary was similar across the two groups, whereas the correlation with narrative content was only significant in the typically developing group. When age and nonverbal ability were partialled out, the general pattern of results and the significant differences between the two groups remained. The most notable difference from the simple correlations was that the relationship between nonword reading and reading comprehension was no longer significant for the children with Down syndrome.

Reading comprehension at Time 1 was then controlled for. The correlations largely reduced and few were significant for the typically developing group. The exceptions were receptive vocabulary, narrative content and digit recall. Reading accuracy was not significant in either group, presumably because this had high amounts of shared variance with reading comprehension at Time 1. For the children with Down syndrome, phonological skills were significantly correlated with reading comprehension, as were the two memory measures, receptive vocabulary and MLU. In the case of block recall and MLU, the correlations with reading comprehension for the children with Down syndrome were significantly higher than for the typically developing group.

Path models were conducted with reading comprehension at Time 3 as the outcome measure and reading accuracy (Read1) and the vocabulary composite at Time 1 (Vocab1), along with reading comprehension at Time 1 (Comp1), were entered as predictors. These variables were chosen to evaluate the relative contributions of reading accuracy and oral language to growth in reading comprehension. These path models are shown in Figure 6. For the typically developing children, the pathways from initial reading accuracy and vocabulary to later reading comprehension were significant, although for the children with Down syndrome, only the path from vocabulary to reading comprehension was significant. The difference in the strength of the pathway from reading accuracy to reading comprehension between the two groups was marginally significant (p=.056).

In summary, despite being matched on reading accuracy skills at Time 1, the children with Down syndrome performed significantly more poorly on reading comprehension than the typically developing children at all time points. For both groups, word and nonword reading were similarly correlated with reading comprehension, although in the path models with reading comprehension, reading accuracy and vocabulary as predictors, reading accuracy only predicted unique variance in reading comprehension for the typically developing children. Vocabulary accounted for unique variance in both groups, whereas initial reading comprehension was not significant in either group. It is also worth noting that in the longitudinal correlations, both MLU and digit recall were significantly more strongly correlated with reading comprehension for the children with Down syndrome than the typically developing children.

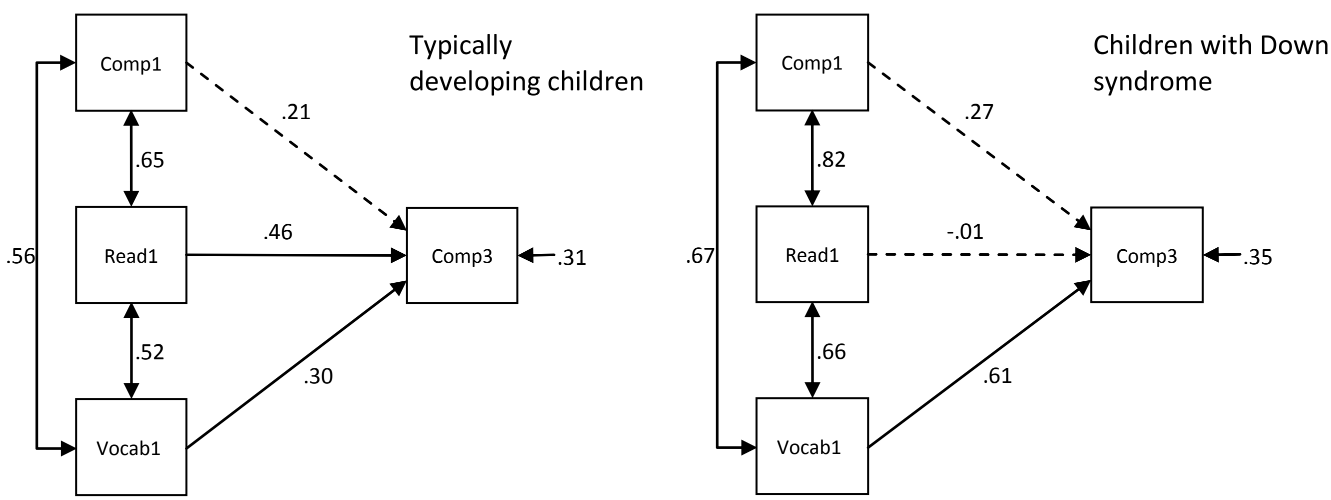


Figure 6. Path models using simultaneous regression and residuals standardised for age to predict Time 3 reading comprehension with Time 1 reading comprehension, reading accuracy and vocabulary as predictors (Study 1)

2.3.7.3. Predicting oral language from reading accuracy.

The correlations between reading accuracy at Time 1 and later oral language measures are shown in Table 16. The oral language measures were taken from Time 3 if they were included in the test battery, or Time 2 if not. Considering the phonological skills first, the correlation with alliteration matching was moderate in the children with Down syndrome but not significant. As discussed previously, phoneme deletion is a more sensitive task of phonological awareness than alliteration matching for the typically developing group and this was highly correlated with earlier reading accuracy. Earlier reading accuracy was significantly correlated with nonword repetition in the children with Down syndrome only. The correlations with both vocabulary measures were strong and significant in both groups. The relationship of reading accuracy with sentence structure, MLU and narrative content was slightly higher and therefore significant only in the typically developing group.

Partial correlations controlling for Time 1 age and nonverbal ability were also conducted (Table 16). Generally the strength of the partial correlations reduced and many were no longer significant. Reading accuracy was still significantly correlated with vocabulary in both groups, and also with phoneme deletion in the typically developing group.

Partial correlations controlling for the autoregressive effect are also shown in Table 16. The earliest administration of the task was used, which was at Time 1 with the exception of phoneme deletion that was first administered at Time 2. After partialling out the autoregressor, reading accuracy was only significantly correlated with picture naming in the typically developing group, although there was a similarly moderate relationship with BPVS in the children with Down syndrome.

In summary, previous research has suggested that learning to read may promote oral language development in Down syndrome. There are also studies that suggest reading accuracy may have an impact on future phonological skills in typical development. Therefore the correlations between Time 1 reading accuracy and later oral language measures were examined. The most robust results are for vocabulary in both groups and phoneme deletion in the typically developing group, although only the correlation with picture naming in the typically developing group remained significant after the autoregressors were controlled for.

Table 16.
Correlations of Time 2 and Time 3 oral language measures with Time 1 reading accuracy for the children with Down syndrome and the typically developing children (Study 1)

	Simple correlations		Partial correlations controlling for Time 1 age and matrices		Partial correlations controlling for the autoregressor	
	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children
T3 Alliteration matching	.45	.03	.32	.19	.12	-.04
T3 Phoneme deletion	-	.64**	-	.60**	-	.22
T2 Nonword repetition	.50*	.22	.40	.13	.17	-.08
T3 Picture naming	.65**	.70**	.57*	.64**	.15	.48**
T3 BPVS	.64**	.69**	.54*	.44*	.41	.32
T3 Vocabulary composite	.64**	.72**	.55*	.59**	.16	.37[a]
T2 Sentence structure	.41	.57**	.13	.11	-.02	.22
T3 MLU	.36	.40*	.26	.19	.11	.25
T3 Narrative content	.22	.46*	-.05	.10	.11	.36

Note. [a]p=.051
*p<0.05 **p<0.01

2.4. Discussion

The aim of Study 1 was to examine the interrelationships between reading accuracy, reading comprehension and oral language development in children with Down syndrome. It was hypothesised that oral language skills would predict reading accuracy to a greater extent than phoneme awareness in children with Down syndrome, whereas the opposite would be true for typically developing children. In longitudinal path models, initial levels of reading accuracy predicted Time 3 reading accuracy for both groups of children but due to the strong autoregressive effect, phoneme awareness and vocabulary did not. In concurrent path models, vocabulary but not phoneme awareness predicted reading accuracy for the children with Down syndrome. Despite being significantly correlated with concurrent reading accuracy, neither vocabulary nor phoneme awareness added unique variance to reading accuracy in the typically developing children, although there was a trend in the results for phoneme awareness to be a significant predictor. Therefore results support the prediction made for the children with Down syndrome, but the results for the typically developing children are mixed.

The relative contribution of oral language and reading accuracy to reading comprehension was evaluated in line with the simple view of reading. It was expected that the relationships would be similar across the two groups at the first time-point, with reading accuracy playing a prominent role in predicting reading comprehension. This was indeed found to be the case. Longitudinally it was predicted that a similar pattern should remain for the children with Down syndrome, whereas there would be a more equal contribution from both oral language and reading accuracy for the typically developing children due to greater progress in literacy development. This prediction was supported for the typically developing children but unexpectedly only oral language predicted reading comprehension for the children with Down syndrome.

The effect of reading accuracy on later oral language skill was also investigated. It was hypothesised that reading accuracy would predict phonological skills in both groups and the extent of this was to be compared between the two groups. The aim regarding broader oral language skills was more exploratory. There was some evidence that reading accuracy predicted later vocabulary skills in both groups and phoneme deletion in the typically developing group. When controlling for the autoregressor, only the relationship with picture naming in the typically developing group was significant although the correlation with receptive vocabulary was a similar strength for the children with Down syndrome.

2.4.1. Reading Accuracy

2.4.1.1. Reading as an island of ability in Down syndrome

An uneven cognitive profile is associated with Down syndrome, with a general advantage for visual compared to verbal skills. Reading accuracy is reported to be a strength, although the extent of this has been disputed (Cardoso-Martins et al., 2009). At Time 1 the groups were matched on reading accuracy but the typically developing group performed significantly better than the children with Down syndrome on tasks of nonverbal ability, reading comprehension, nonword reading, phonological skills, broader oral language and memory. Therefore reading accuracy was a clear strength in this group of children with Down syndrome relative to all other cognitive domains assessed, supporting Buckley's (1985) concept of reading accuracy as an island of ability. As found in previous longitudinal studies (e.g. Byrne et al., 2002; Hulme et al., in press), there was less progress in reading accuracy in the children with Down syndrome than the typically developing children over the course of study; the average gain for the children with Down syndrome was nine months compared to two years for the typically developing group.

2.4.1.2. Phoneme awareness, letter-sound knowledge and the alphabetic principle.

Phoneme awareness is a key longitudinal predictor of reading accuracy in typically developing children (e.g. Muter et al., 2004) but not for children with Down syndrome (Hulme et al., in press). In the present study phoneme awareness was not a unique predictor of the development of reading accuracy for the children with Down syndrome, but more unexpectedly neither was it for the typically developing children. However there was a large degree of longitudinal stability in reading accuracy in both groups, as illustrated by the strong autoregressive effects in the path models, which left little variance remaining especially for the children with Down syndrome.

Autoregressive effects are problematic when there is such a high level of stability (e.g. Chan, 2003). Therefore it is important to consider what variables relate to initial levels of reading accuracy, i.e. at Time 1. Path models examined this with phoneme awareness and vocabulary entered as predictors. Phoneme awareness did not significantly predict reading accuracy in the children with Down syndrome, although it should be noted that the strength of the correlations between the two skills were weak to moderate suggesting some degree of association. In the concurrent correlations phoneme awareness was related to reading accuracy for the typically developing group as would be expected and there was a trend for phoneme awareness to predict reading accuracy in the path model.

The causal relationship between phoneme awareness and reading accuracy is an established finding in studies with typically developing children (e.g. Bradley & Bryant, 1983; Muter, 1994; Muter et al., 2004; Stuart & Masterson, 1992; Wagner et al., 1997); therefore the non-significant finding was unexpected for the typically developing children. There are a number of reasons why this may have occurred. Firstly, as the children ranged in age, controlling for this variable in the path models may have removed a large proportion of the variance. Secondly, the phoneme awareness task used, alliteration matching, may have had a relatively high load on vocabulary, leading to shared variance. Indeed the correlation between vocabulary and alliteration matching was significant. If a child was familiar with the items in the alliteration matching task then they would have had a pre-existing phonological representation that may have made it easier to identify the initial sound. Thirdly, the typically developing children tended to score quite well on the alliteration matching task, and its lack of sensitivity and narrow range of scores may have affected the potential of the task to predict variance in reading. This task was adapted from a study with typically developing children at the early stages of phonological awareness development (Carroll, 2004) and alliteration matching tasks have been previously used with children with Down syndrome (Kennedy & Flynn, 2003; Roch & Jarrold, 2008). Therefore it was thought this task would be of an appropriate difficulty level for the children with Down syndrome, however this resulted in it being too easy for the typically developing children, whose scores were close to ceiling.

Along with phoneme awareness, letter-sound knowledge is considered critical for the development of reading accuracy in typically developing children due to its role in the alphabetic principle (Byrne & Fielding Barnsley, 1989). In the concurrent and longitudinal correlations, letter-sound knowledge was significantly related to reading accuracy in the children with Down syndrome. This is in contrast to previous research which has found that letter-sound knowledge does not correlate with reading accuracy in children with Down syndrome (Boudreau, 2002; Snowling et al., 2002). It may be that reading instruction is now focussing more on letter-sound knowledge, a task on which children with Down syndrome can perform relatively well. Indeed in a more recent study, letter-sound knowledge was also found to be moderately correlated with reading accuracy in children with Down syndrome (Hulme et al., in press).

The children with Down syndrome had relative weaknesses on letter-sound knowledge and phonological awareness, which suggest potential problems with the acquisition of the alphabetic principle and this would be likely to underlie their relative difficulties on the nonword reading task (see also Hulme et al., in press). A stronger test of the alphabetic principle would be to use nonword reading as an outcome measure. However even at Time 3 in this study, a third of the children with Down syndrome scored zero and another third scored under five on this task, therefore variability was low and results would be skewed. The nonword reading task used in this study began with nonwords which were four letters long; to increase the sensitivity of this task in future studies, two or three letter nonwords should be added. Furthermore a task of phoneme blending should be included, as this would represent the assembly process which is thought to be necessary for decoding along with the alphabetic principle (Byrne & Fielding-Barnsley, 1989).

Despite the discrepancy between word and nonword reading in the children with Down syndrome, the correlation between word and nonword reading was similar in the two groups at all three time points, suggesting that regardless of group membership, children who had higher levels of reading accuracy were also better at reading nonwords, i.e. the rank-order was similarly stable across the two tasks in the two groups.

2.4.1.3. Broader oral language skills.

A key aim of this study was to evaluate the contribution of a broad range of oral language measures to reading accuracy, therefore tasks of receptive vocabulary, picture naming, receptive syntax, MLU, narrative content and nonword repetition were included. The results for each of these will now be discussed in turn.

It was the vocabulary measures in both groups that were most consistently related to reading accuracy across all the time-points and therefore this was chosen to index oral language in the path models. When the Time 1 vocabulary composite was entered into the path model to predict Time 3 reading accuracy, along with phoneme awareness and reading accuracy, it was not a unique predictor in either group due to the strength of the autoregressor. Concurrent path models for Time 1 with phoneme awareness and vocabulary as predictor variables showed that vocabulary was a significant predictor of reading accuracy for the children with Down syndrome but not for the typically developing children.

Vocabulary knowledge consists of both lexical phonology, i.e. familiarity with the phonological forms of words, and semantics, i.e. knowledge of the meanings of words (Nation & Cocksey, 2009). The tasks used in this study can be considered measures of the breadth of vocabulary, which assess the phonological representation of words but also incorporate shallow semantic knowledge (Ouellette, 2006). Ouellette found that measures of vocabulary breadth were correlated with irregular word and nonword reading for typically developing children and one of the reasons suggested for these relationships was the common processes of learning, storing and retrieving phonological representations.

Traditional measures of vocabulary breadth may tap phonological representations more than semantics but the two components cannot be separated. Using a lexical decision task to assess phonological familiarity without semantic interference and a definitions task to represent semantics, Nation and Cocksey (2009) found that lexical phonology predicted reading accuracy rather than semantics. The words used in Nation and Cocksey's study were irregular, and the authors argued that familiarity with the phonological form of the word allows children to produce the correct word on the basis of a partial decoding attempt. Similarly, vocabulary knowledge may predict reading accuracy in children with Down syndrome by providing a corresponding phonological representation that supports partial decoding attempts. This may be more important than for the typically developing children as the children with Down syndrome had poorer nonword reading and therefore are likely to produce more partial decoding attempts.

It has been argued that lexical phonology underlies the relationship between vocabulary knowledge and reading accuracy, and this will be returned to in Chapter 5. However it is important to note that phonological and semantic knowledge cannot be separated in the present vocabulary tasks and it could be that semantics also plays a role. Indeed Snowling, Gallagher and Frith (2003) suggested that children with poor phonological processing but age-appropriate reading accuracy were able to compensate for their relatively inefficient phonological processing using their oral language skills, i.e. via semantic and sentence contexts.

The measures of MLU, receptive syntax, narrative content and nonword repetition and their relationship with reading accuracy concurrently at Time 1 and longitudinally will now be considered. MLU was correlated more strongly with reading accuracy concurrently and longitudinally for the children with Down syndrome than the typically developing children, although there was some indication that this relationship may operate via vocabulary. In contrast, a receptive task of grammar was significantly correlated with reading accuracy for only the typically developing children at Time 1 and longitudinally. Receptive syntax tasks may aid reading accuracy by providing syntactic constraints on a possible word. However this task was not correlated with reading accuracy for the children with Down syndrome, possibly because they have lower levels of syntactic knowledge and therefore are less able to utilise this skill when reading. Nonword repetition was correlated with reading accuracy at Time 1 and longitudinally in both groups. However this was no longer the case when the autoregressor was controlled for and there is recent evidence to suggest that this relationship may primarily operate in the reverse direction (Nation & Hulme, 2011). Narrative content was related to reading accuracy concurrently for the typically developing children but not the children with Down syndrome, supporting the findings of Boudreau (2002). However narrative content did not appear to have a robust longitudinal relationship with reading accuracy in either group.

2.4.2. Reading Comprehension

Reading comprehension is typically a weakness in comparison to reading accuracy in children with Down syndrome (e.g. Nash & Heath, 2011). It was expected that this finding would be replicated here, with the children with Down syndrome performing significantly below their reading accuracy matched control group on reading comprehension at Time 1. This was confirmed and reading comprehension was delayed by an average of 11 months relative to reading accuracy in the group of children with Down syndrome.

An aim of this study was to evaluate the simple view of reading in children with Down syndrome. According to the simple view of reading, reading comprehension is the product of being able to read accurately and understand spoken language, although the relative contribution of these two skills differ throughout development depending on a child's level of reading accuracy (Vellutino et al., 2007). At Time 1, it was expected that reading comprehension in both groups would be more reliant on reading accuracy than oral language, which was confirmed by a path model. When Time 1 reading comprehension, reading accuracy and vocabulary were entered as predictors of later reading comprehension, reading accuracy only predicted unique variance for the typically developing children. Vocabulary accounted for a similar amount of unique variance in reading comprehension in both groups.

The finding that reading accuracy did not relate to later reading comprehension in the group of children with Down syndrome was unexpected. Indeed the beta weights suggested that reading accuracy had very little effect on later reading comprehension, which seems unlikely. As the groups were not matched on passage reading accuracy at Time 3 the reading comprehension scores for the children with Down syndrome were calculated from earlier passages in the YARC passage reading test. This is important as the first passage in the YARC is a shared reading task in which the experimenter and child read alternate lines, therefore this also taps listening comprehension, which would place a greater load on oral language than reading accuracy.

In the longitudinal and concurrent correlations, MLU and verbal short-term memory were significantly related to reading comprehension for the children with Down syndrome but not for the typically developing children. The reading comprehension task used in this study required children to answer open-ended questions, therefore putting demands on their ability to produce spoken sentences. As children with Down syndrome have language production difficulties, and therefore low MLUs, it is suggested that this may constrain how well they can express their understanding and thus could limit performance on reading comprehension tasks. Working memory is one of several higher-level processes that have been suggested to impact on reading comprehension (Kintsch & Rawson, 2005; Perfetti et al., 2005). As this is typically a weakness for individuals with Down syndrome, they may have difficulty forming and holding a situation model of the text in memory and will therefore have difficulty answering comprehension questions requiring the synthesis of information across the text.

2.4.3. Oral Language

This study aimed to investigate the effect of reading accuracy on oral language development. Firstly the effect on phonological skills will be considered. Reading accuracy was not related to later nonword repetition in either group but there was a significant correlation with later phoneme deletion in the typically developing group although this was no longer the case when the autoregressor was controlled. In contrast, reading accuracy has been previously found to predict later nonword repetition (Nation & Hulme, 2011) and phoneme awareness (Castro-Caldas et al., 1998; Hulme et al., in press; Morais et al., 1979) in typically developing individuals. Previous results are mixed for children with Down syndrome, with Hulme et al. (in press) finding a significant relationship between reading accuracy and later phoneme awareness even when accounting for the autoregressor whereas Cupples and Iacono (2000) did not. Therefore there is some conflict between the present results, particularly for typically developing children, and those of previous studies and findings clearly need replicating. Possible reasons for these different results may be due to the different nonword repetition and phoneme awareness tasks used and the differences in sample size and therefore statistical power.

Initial reading accuracy was significantly correlated with Time 3 vocabulary in both groups even when partialling out age and nonverbal ability. When the autoregressor was included, only the correlation with picture naming in the typically developing group remained significant, although the relationship with receptive vocabulary was still moderate in the children with Down syndrome. Learning to read exposes children to more opportunities to acquire new vocabulary and is also thought to promote segmental representations (Ziegler et al., 2003), which may then result in more accurate phonological representations of object's names.

The argument that reading accuracy promotes language development in Down syndrome mainly stems from case studies (e.g. de Graaf, 1993; Duffen, 1976) and there is the implication that this is because of their unique cognitive profile (Buckley & Bird, 1993) and therefore this would be expected to be a stronger effect than in typical development. The present study suggests that this is not the case.

2.4.4. Limitations

The children with Down syndrome were matched to the typically developing children on reading accuracy at the first time-point. This presents two issues: firstly, the groups were not matched on any other variable; secondly, there was a different rate of development in the two groups. It was expected that the children with Down syndrome would show weaknesses relative to the control group on tasks other than reading accuracy. This meant that the test battery needed to incorporate tasks that would cover a range of abilities and in two instances this resulted in the use of slightly different versions of a task, and therefore age-equivalent scores were used in the analyses which can be problematic (Mervis & Klein-Tasman, 2004). Furthermore on a number of tasks, most noticeably alliteration matching, ceiling effects were present for the typically developing children whilst the children with Down syndrome found this task relatively difficult. Due to their learning difficulties, the children with Down syndrome made less progress than the typically developing children during the course of the study. This resulted in different levels of reading accuracy at the end-point and therefore different amounts of progress being predicted in the analyses. Both these issues are inherent when working with children with learning difficulties and uneven cognitive profiles, and should be borne in mind when making comparisons with typically developing children. The most pertinent consequence in the present study is that 88% of variance in Time 3 reading accuracy in the children with Down syndrome is accounted for by initial levels of reading accuracy, compared to 69% for the typically developing children. This renders it unlikely that any other remaining variable would predict unique variance in samples of the size studied here.

Typically studies use 50-100 utterances to calculate MLUs (Eisenberg, Fersko, & Lundgren, 2001) but in this study the total number of utterances produced was relatively low, with means ranging from 6.67 to 10.37 across groups and time-points. Therefore the MLUs computed may be unreliable, especially for the children with Down syndrome for whom the variability was larger. In other studies with individuals with Down syndrome, the total number of utterances has been similar to the present study in narrative contexts (Boudreau & Chapman, 2002), whereas others have collected more utterances by requiring individuals to narrate multiple picture books or also talk about family photos (Miles & Chapman, 2002; Miles et al., 2006). However a recent study found strong correlations between MLUs calculated using ten utterances and larger language samples (which had a mean of 114 utterances) for children with language impairment (Casby, 2011), suggesting MLUs can be reliably calculated from short language samples.

2.5. Conclusions

Reading accuracy was found to have high levels of stability over time in both groups but particularly so for the children with Down syndrome. In a path model investigating longitudinal predictors of reading accuracy, this led to strong autoregressive effects and neither phoneme awareness nor vocabulary added unique variance. This was the case for both groups. When concurrent path models were conducted with vocabulary and phoneme awareness as predictors, vocabulary was a unique predictor of reading accuracy for the children with Down syndrome whereas phoneme awareness was not. Neither measure uniquely predicted reading accuracy in the typically developing group although it is important to note that there was a trend for phoneme awareness to be significant. In summary, the pattern of results were as expected for the children with Down syndrome, although it should be noted that the relationship between phoneme awareness and reading accuracy was weak but it was not non-existent nor was it significantly weaker than in the typically developing children.

The simple view of reading was evaluated, and longitudinally oral language predicted reading comprehension in both groups of children but reading accuracy did not predict reading for the children with Down syndrome, although this may be for methodological reasons. At Time 1, when the groups were matched on reading accuracy, a clearer picture emerged. At this early stage in literacy development, reading accuracy was a unique predictor of reading comprehension for both groups but oral language was not. MLU, a measure of expressive sentence complexity, and verbal short-term memory were more strongly related to reading comprehension in children with Down syndrome than in typically developing children and this may relate to the memory and expressive language demands required to complete the reading comprehension task.

The effect of reading accuracy on oral language development was also examined. Initial levels of reading accuracy were moderately related to vocabulary development in both groups, although this was not significant for the children with Down syndrome. There was some evidence that reading accuracy promoted phoneme awareness in the typically developing group, but this was not significant when employing the rigorous control of the autoregressor. Therefore this suggests that reading accuracy may promote the development of vocabulary but this requires replication and that the effect is not unique to individuals with Down syndrome.

Chapter Three

The Benefit of Orthographic Support for Spoken Word Learning in Children with Down Syndrome

3.1. Introduction

3.1.1. Overview

Oral language in children with Down syndrome has typically been examined by assessing acquired language skills concurrently and longitudinally. This is useful as it can provide information on the level of language ability attained and its relationship with other skills. Dynamic tasks of language learning complement this approach well by providing an insight into the underlying processes and rate of language acquisition.

This chapter will first review existing studies of spoken word learning in children with Down syndrome and then discuss the proposal that reading can promote language development. Experimental studies with typically developing children have examined the effect of orthographic support when teaching new spoken words and these will be described. Study 2 will then be presented, which adopted a similar paradigm and compared the benefit provided by orthographic support to the phonological learning of new spoken words in children with Down syndrome compared to typically developing children.

3.1.2. Fast-Mapping Studies with Individuals with Down Syndrome

The fast-mapping paradigm has been used with individuals with Down syndrome to investigate spoken word learning. Fast-mapping is a form of incidental learning where a novel label for a novel object is introduced in the context of another task, often a game (Carey & Bartlett, 1978). Probes can be used during the training (or exposure) phase that assess children's ability to comprehend or produce the target word, for example asking the child to pick up the target object or asking them what it is called. Children's spontaneous imitations of the word during the training phase can also be assessed. The comprehension and production tasks can also be conducted as post-tests.

The first fast-mapping study carried out with individuals with Down syndrome was by Chapman et al. (1990) with 48 individuals with Down syndrome aged 5-20 years and 48 typically developing children aged 2-6 years who were matched on nonverbal ability. Children were exposed to a novel object-word mapping within the context of a game and then took part in immediate and one hour delayed comprehension and production tasks. In each group most children succeeded on the comprehension task and approximately half produced the correct word. The groups also performed similarly on a task of existing receptive vocabulary, which suggests that children with Down syndrome perform in line with both their nonverbal ability and acquired vocabulary knowledge on a task of spoken word learning. However as Mervis and Bertrand (1995) highlight, Chapman et al. did not address whether children had acquired the novel name-nameless category (N3C), which is the ability to fast map a novel word to a (basic-level) category for which they do not already have a label. Mervis and Bertrand found that nine out of 22 children with Down syndrome aged 2-3 years were able to acquire this principle, which was defined as mapping the novel label to the novel object and generalising this to other examples of the same category. Importantly these children had larger vocabularies than the 13 children with Down syndrome who were not able to acquire the principle, suggesting the acquisition of the N3C principle is related to vocabulary spurts in early development as for typically developing children.

Fast-mapping can also be carried out in the context of a story, as in Kay-Raining Bird, Chapman and Schwartz's study (2004) with a subsample of the older participants from Chapman et al. (1990). Twenty-three individuals with Down syndrome aged 12-20 years and 24 typically developing children aged 4-6 years matched on nonverbal ability took part. Participants heard four stories, each containing two unique novel words, the meaning of which mapped onto an existing word. After hearing the story, the participants were asked to retell the story and then define the novel words. The number of spontaneous productions of the novel word within the story retelling was used as the measure of fast-mapping production. A similar number of children in each group produced the novel word at least once whilst retelling the story, and overall the number of successful productions did not differ across groups. There was a trend for individuals with Down syndrome to define fewer words than the typically developing children but this was not significant. The overall findings of this study support that of Chapman et al. (1990) suggesting that individuals with Down syndrome have fast-mapping abilities in line with their nonverbal ability.

The fast-mapping studies described above have only investigated the fast-mapping of nouns. McDuffie, Sindberg, Hesketh and Chapman (2007) extended this work by investigating the fast-mapping of nouns along with verbs with 20 individuals with Down syndrome, aged 12-18 years, and 19 typically developing children, aged 3-6 years, matched on receptive syntax. The paradigm employed was similar to the classic fast-mapping game but in the context of a magic show. The ability to correctly name the novel object was very low across groups and word types. On a comprehension task, the two groups performed similarly and both found verbs more difficult than nouns. Therefore children with Down syndrome are similar to typically developing children in finding production more difficult than comprehension and verbs more difficult to learn than nouns. The matching criterion of receptive syntax resulted in higher scores for nonverbal ability, and there was also some evidence of higher scores for receptive vocabulary in the individuals with Down syndrome. Therefore children with Down syndrome had equivalent fast-mapping ability to children of a *lower* nonverbal ability, in contrast to Chapman et al. (1990) and Kay-Raining Bird et al. (2004)

Children with Down syndrome are often taught some form of sign language at a young age, for example makaton. Therefore Kay-Raining Bird, Gaskell, Dallaire and MacDonald (2000) compared fast-mapping ability in young children with Down syndrome in both spoken and signed modalities. Ten children with Down syndrome aged 2-5 years and a group of 10 typically developing children aged 1-2 years took part. The groups were matched on a parental report of expressive vocabulary and the Bayley scales, which incorporates nonverbal, language and motor skills. Novel objects were labelled with a novel word, a novel sign or both. Children with Down syndrome spontaneously imitated more than the typically developing children, and both groups imitated most in the combined condition, where it was the word rather than sign that was mostly produced. On a comprehension task the typically developing children performed better than the children with Down syndrome. Comparing across conditions, the children with Down syndrome comprehended more words in the combined condition whereas the typically developing children performed similarly in all conditions. This suggests that the imitation of a new spoken word is more likely with dual modality presentation, and this may aid spoken word comprehension in Down syndrome to a greater extent than typical development. However this finding needs replication as the groups were not equated for familiarity to signing and the small sample size precluded the use of statistical analyses.

The number of repetitions of the target nonword during a fast-mapping task can be varied to examine whether increased presentations improve new word learning. Chapman, Sindberg, Bridge, Gigstead and Hesketh (2006) presented a nonword once or four times to 19 adolescents with Down syndrome aged 12-21 years, and 18 typically developing children aged 2-5 years matched on receptive syntax. The groups also had similar levels of receptive vocabulary but the children with Down syndrome had higher nonverbal ability. Four repetitions of the nonword increased accuracy on the comprehension tasks for the typically developing children and speed of response on the comprehension tasks for the children with Down syndrome. The accuracy of production for the nonword was also increased in both groups. There were no overall differences between the two groups on the comprehension and production measures of accuracy but the typically developing children were quicker overall. Therefore the children with Down syndrome performed largely in line with their receptive language level, and generally both groups benefited from hearing the target word more.

Chapman et al. (2006) also tested the effect of eliciting production of the nonword during the fast-mapping task. When children were required to say the new word during training, this benefited the production of the nonword of the children with Down syndrome who had poor hearing. There were no effects on comprehension during training. This suggests that for typically developing children and children with Down syndrome with unimpaired or corrected hearing levels, eliciting production of a nonword does not help to learn it, but it may help children with Down syndrome who have hearing difficulties.

The fast-mapping studies appear to show that children with Down syndrome can learn new spoken words and their referents to the same level as typically developing children matched on nonverbal ability and/or receptive language. However all these studies have been carried out by the same research group and with the exception of Kay-Raining Bird et al.'s (2000) study on signing, all participants were recruited for the same geographical area. Indeed Kay-Raining Bird et al. (2004) state that their sample originally took part in Chapman et al.

(1990). Therefore some of the same participants will have taken part in multiple studies, causing similar findings to be more likely.

The production tasks used in the fast-mapping studies were lenient in what was accepted as a correct answer; a response was still considered correct if there was an error on one phoneme in the target word, or if a phoneme was added. This may disguise differences between the groups, for example in Kay-Raining Bird et al. (2004) the children with Down syndrome were most likely to produce an approximate spoken response, whereas the typically developing children were most likely to produce an accurate spoken response, although both were considered correct. In comprehension tasks, the names of the other objects were often acoustically dissimilar, e.g. ‘pen’ and ‘spoon’ in Chapman et al. (1990), and therefore the comprehension task, along with the production task, could be successfully completed even if the child had a relatively poor phonological representation of the novel word.

3.1.3. Assessing the Quality of Phonological Representations in Individuals with Down Syndrome

Problems with articulation are common in children with Down syndrome (Kumin et al., 1994; Roberts et al., 2005), thus making it difficult to assess the production of new words. To circumvent this whilst stringently assessing the quality of phonological representations, Jarrold et al. (2009) tested spoken word learning using a receptive multiple choice task where the distracters were phonetically similar to the target nonwords. The rationale was that if children had poor phonological representations they would not be able to successfully discriminate between the target nonwords and distracters. Nonwords were presented as the names of aliens to 22 individuals with Down syndrome aged 14-29 years and 64 typically developing children aged 5-8 years. The groups were not explicitly matched on any measure but performed similarly on tasks of nonverbal ability and vocabulary knowledge. After the presentation phase, in which three aliens were shown individually on a computer screen and named, children’s phonological and referent learning was tested. To test phonological learning, individuals were presented with a picture of an alien and had to pick the correct name out of a choice of three spoken nonwords. To test referent learning, participants heard a name and had to choose the matching alien from a choice of three. The two groups performed equivalently on the referent learning task, but the group of individuals with Down syndrome performed significantly worse than the typically developing group on the phonological learning task and this difference was accounted for by performance on verbal short-term memory tasks. Thus it appears that when assessed more strictly, individuals with Down syndrome have specific problems with learning the phonological forms of new words and this is directly linked to their deficit in memory.

Mosse and Jarrold (2011) extended this work by using a similar training methodology but requiring a spoken response. Seventeen individuals with Down syndrome, aged 9-28 years, and 24 typically developing children, aged 4-6 years and of a similar nonverbal and vocabulary ability, took part. In the training phase individuals were presented with three boys and their names on a computer. In the testing phase, children had to produce the name of each boy. The names were familiar e.g. Simon, and there was also a nonword version of the task with robots who had nonwords for names. There were no significant differences between the two groups and both showed a lexicality effect with an advantage for words over nonwords. In a second experiment, the wordlikeness of nonwords was varied and a group of individuals with Down syndrome performed equivalently to a group of typically developing children and both groups performed significantly better with nonwords that were word-like. When z-scores of verbal short-term memory and the word and nonword learning tasks were calculated based on the typically developing children's scores, the children with Down syndrome showed a significant discrepancy, i.e. their word and nonword learning was better than one would predict based on their verbal short-term memory. The authors explained this finding by suggesting that individuals with Down syndrome rely less heavily on their verbal short-term memory and more on a domain-general serial order processing mechanism, which supports their word learning by the repetition of information.

The findings of Mosse and Jarrold (2011) suggest that individuals with Down syndrome show phonological learning in line with their nonverbal ability and vocabulary skills which supports the fast-mapping literature but is in contrast to Jarrold et al. (2009). To attempt to reconcile this, Mosse and Jarrold conducted a third experiment with a receptive multiple-choice task which aimed to replicate Jarrold et al. However the children with Down syndrome did not show impairments in phonological learning relative to the typically developing children.

Although this study aimed to replicate Jarrold et al. there were some differences in terms of participants and methodology. In Mosse and Jarrold the typically developing group were five years old compared to six years old in Jarrold et al.. Furthermore the distracters and target items were equated for presentation frequency, whereas as in Jarrold et al. the distracters were presented fewer times than the target item. Mosse and Jarrold argued that the reason for the discrepancy in the results is that in Jarrold et al., the typically developing children may have been more likely to choose the target item based on familiarity.

3.1.4. The Proposed Benefit of Reading Accuracy on Vocabulary Learning in Down Syndrome.

It is has been proposed that reading accuracy benefits oral language in children with Down syndrome (e.g. Buckley, 1995) and longitudinal studies relating to this were reviewed in Chapter One. Here reasons why seeing the orthographic form of a word may help children with Down syndrome to learn its spoken form will be briefly considered.

Case studies with children with Down syndrome have argued that presenting a word in its orthographic form promotes the use of this word in speech (de Graaf, 1993; Duffen, 1976). There are, at least, two reasons why this may occur. Buckley (1995) argued that “reading practice improves phonology and articulation, possibly because the letters in words provide the cues the child needs to sound all the phonemes” (p. 161). If children can identify the individual phonemes in a new spoken word then this may result in their phonological output, and therefore representation, being more accurate. Additionally orthography may provide children with another representation of the new word form, which strengthens the overall representation in the lexicon and therefore aids retrieval at a later date. This is in line with Perfetti and Hart’s (2002) lexical quality hypothesis, which states that a lexical representation is high quality when it has fully specified orthographic, phonological and semantic representations.

Buckley (1993) compared grammatical learning with and without supporting text with 12 adolescents with Down syndrome, aged 12-15 years. Children saw a picture depicting an action and heard an accompanying sentence. For half of the sentences, this was also presented in its written form. Children then had to say the sentence with only the picture present. When the sentences were seen by the individual during training they were better able to independently produce the sentence. Buckley argued that seeing the written word resulted in its visual image being stored in memory and this then helped recall, supporting Perfetti and Hart's (2002) lexical quality hypothesis.

Although the empirical evidence is limited in quantity, it is plausible that due to their relative strengths in the visual domain and reading accuracy compared to weaknesses in oral language, the spoken word learning of children with Down syndrome may benefit from orthography. However, this has not yet been tested outside of case studies or in comparison to typically developing children.

3.1.5. The Effect of Orthography on Spoken Word Learning in Typically Developing Children

There have been studies with typically developing children which have directly examined whether seeing the orthography of a word helps children to learn its phonology and semantics. The first of these, Ehri and Wilce (1979), taught spoken nonwords paired with visual stimuli to children aged six to eight years. There were different types of visual stimuli: a squiggle, the initial letter of the word only, the initial letter with a misspelling of the word and the initial letter with the correct spelling of the word. When the correct spelling was presented, learning was significantly faster than in the other three conditions. These results indicate that seeing the correct spelling of a word helps children learn its phonological form, and this effect is specific to orthography rather than just a general visual cue. However this study is limited by its representation of word learning. The nonwords were paired with squiggles or letters rather than objects or definitions, and as such, contained no semantic information and did not resemble word learning in everyday life.

More recently, there have been two studies that directly assessed the effect of orthography on the phonological and semantic aspects of word learning. Rosenthal and Ehri (2008) adapted Ehri and Wilce's (1979) study to teach 12 low-frequency nouns along with their definitions and pictures, six of which were taught with their orthography also present. During training children were required to recall the word when they saw the picture and recall the definition when they heard the word. One day after the training, children took part in three post-tests: producing the spoken word when the definition was heard, producing the spelling when the word was heard and matching the novel words with meaning-elaborating sentences.

For words which were trained with orthography, children were quicker to learn the pronunciations and meanings during training and were more accurate on the spelling post-test and the pronunciation recall post-test. There was a slight advantage for words taught with their orthography on the matching task, but children performed close to ceiling in both conditions. This first study was carried out with children aged seven to eight years and a second experiment extended the results to children aged 10-11 years. These studies show that providing orthography during spoken word learning aids the learning and recall of both the phonological and semantic representations of the new word. It is suggested that this occurs because children undertake spontaneous and implicit orthographic mapping and link the graphemes in the spelling to the phonemes in the pronunciation, and this then also allows quicker incorporation of the semantic information.

The words used by Ehri and Rosenthal (2008) were based around a synonym, for example a ‘nib’ was described as a pen. Therefore the children may not be learning a new semantic construct, but rather mapping a novel phonological form onto an existing semantic representation, which is more akin to learning a foreign language. Ricketts, Bishop and Nation (2009) also tested the effect of orthography on spoken word learning but used a different training paradigm to Rosenthal and Ehri that trained novel semantic referents. Twelve nonwords were paired with pictures of novel objects and were taught to 58 children aged 8-9 years. Training took the format of three repetition and three production trials, which trained and tested the pronunciation of the nonword and its pairing with the picture. Half of the nonwords were taught with the orthography present and half were taught with the orthography absent. There were two immediate post-tests: spelling and nonword-picture matching. When the nonwords were taught with their orthography, children performed more accurately on the production trials and spelling post-test and were quicker on the nonword-picture matching task. Therefore using a different paradigm, the results from this study support Rosenthal and Ehri’s finding that orthography benefits the learning of phonological and semantic aspects of new word learning.

The effect of orthography on second language learning has also been examined. Hu (2008) taught English nonwords paired with pictures to Chinese-speaking children who had an average age of 8;10. For half the words the orthography was present, and the remaining half were paired with symbols as a control cue. The paradigm was similar to Ricketts et al. (2009) with repetition and production trials. As found in native language learning, children learnt words better when they were paired with their orthography.

In summary, when learning new oral vocabulary, typically developing children with an age range of 7-11 years are benefited by having the orthography present. Specifically it helps them learn the phonology, semantic referent and spelling of the new word. It is argued that the written form of a new word is less transient and variable than its spoken form and creates an orthographic image to represent and reinforce the phonological representation (Ricketts et al., 2009; Rosenthal & Ehri, 2008).

3.1.6. Introduction to Study 2

Individuals with Down syndrome have difficulties in the language domain particularly expressive language (Abbeduto et al., 2007). Studies which have experimentally tested spoken word learning in individuals with Down syndrome have mostly utilised fast-mapping paradigms, and these have found that they demonstrate equivalent levels of comprehension to typically developing children and both groups of children find production tasks more difficult than comprehension tasks (Kay-Raining Bird et al., 2000; McDuffie et al., 2007). Using a different paradigm more akin to paired associate learning it has been found that individuals with Down syndrome can learn referents as well as typically developing children but there are conflicting findings as to whether they are impaired in phonological learning relative to their nonverbal ability and vocabulary knowledge (Jarrold et al., 2009; Mosse & Jarrold, 2011).

It has been proposed that reading accuracy helps the oral language development of children with Down syndrome (Buckley, 1995); this is mostly based on case studies (de Graaf, 1993; Duffen, 1976) and there is mixed support for this from longitudinal studies (Laws et al., 1995; Laws & Gunn, 2002). In studies with typically developing children, experimental paradigms have shown that providing children with the orthography of a new word results in better phonological and semantic learning.

The aim of Study 2 was to examine the learning of new spoken words in children with Down syndrome, and to investigate the effect of orthographic support. Nonwords were paired with a picture of a novel object, as it has been found that both typically developing children and children with Down syndrome find it easier to learn new words when they do not map onto an existing semantic representation (Kay-Raining Bird et al., 2004). To empirically test whether the spoken language of children with Down syndrome can benefit from orthography, and if this is to a greater extent than in typically developing children, half of the nonwords were taught with their orthography present.

Typically developing children were selected to be at a similar reading level to the children with Down syndrome. It was expected that the children with Down syndrome would have poorer existing vocabulary knowledge than the typically developing children and therefore would show lower levels of learning.

It was predicted that the spoken word learning of both groups would benefit from orthography. To ensure any improvement was due to the specific effect of orthography, a control condition which provided a non-orthographic visual cue was included. The relative benefit of children with Down syndrome compared to typically developing children has not yet been investigated and there are two feasible possibilities based on the current literature.

1. Both groups will show a similar advantage from orthographic support because they are of the same reading ability, and therefore have the same opportunity to access and benefit from the written form of the word.

2. Children with Down syndrome may benefit more from orthography than the typically developing children due to their uneven cognitive profile. Their poorer oral language skills coupled with their relatively strong visual skills may result in an additional benefit from orthography.

3.2. Study 2a

3.2.1. Overview

The children with Down syndrome who were to take part in this study were a subsample of those taking part in the longitudinal study, and the vocabulary training studies most similar to the present experiment involved older individuals with Down syndrome (Jarrold et al., 2009; Mosse & Jarrold, 2011). To ensure the learning paradigm would be of an appropriate difficulty level, it was first trialled with typically developing children thought to be of a similar reading ability to the children with Down syndrome who would take part in this study.

3.2.2. Method

3.2.2.1. Design.

Ten novel spoken words were paired with pictures of novel objects. This was a within-participants design, so five nonwords were taught to each child with their orthography present and five were taught with their orthography absent. Performance on production trials during the learning procedure along with a picture naming post-test were the primary outcome measures. A mispronunciation post-test was also administered to test the quality of children's phonological representation of the taught nonwords without requiring a spoken response.

3.2.2.2. Participants.

Sixteen children (seven males) in Year 2 completed this study. They were aged six to seven years, with a mean age of 6;08. Children who had been identified with special educational needs were excluded. Children were recruited from one primary school and consent was gained from both the headteacher of the school and parents.

3.2.2.3. Assessment battery.

3.2.2.3.1. Nonverbal ability.

All children were administered the WPPSI-III^{UK} matrices subtest, which was administered in the same manner as in Study 1 (see section 2.2.3.1.). As all the children completed the same task, in contrast to Study 1, raw scores will be presented.

3.2.2.3.2. Verbal short-term memory.

The word wecall subtest from the WMTB-C was used to measure verbal short-term memory skills. This task was chosen as previous studies of spoken word learning have included memory tasks that use words rather than digits (e.g. Jarrold et al., 2009; Mosse & Jarrold, 2011). The children heard a sequence of words and had to repeat them in the same order. The sequence of words increased in length, until the span score was determined. The number of correct trials, rather than span score, was calculated and used in any analyses. The reliability for this task as reported in the manual is shown in Appendix 1.

3.2.2.3.3. Reading accuracy.

The single word reading test from the YARC Passage Reading test battery was administered. The details of this task can be found in Study 1 (see section 2.2.3.3.3.).

3.2.2.3.4. Phonological awareness.

3.2.2.3.4.1. Alliteration matching.

Alliteration matching was used to assess phonological awareness as in Study 1 (see section 2.2.3.4.1).

3.2.2.3.4.2. Sound deletion.

The sound deletion subtest from the YARC Early Word Reading test battery was also used to assess phonological awareness. Children were presented with spoken words and corresponding colour pictures, asked to repeat the word and then asked to delete a sound (e.g. say sheep without the s). There were 12 items, which tapped deletion of syllables and phonemes in initial, medial and final positions. The reliability for this task as reported in the manual is shown in Appendix 1.

3.2.2.3.5. Expressive vocabulary.

The WPPSI-III^{UK} picture naming subtest was administered as in Study 1 at Time 1 (see section 2.2.3.5.1).

3.2.2.4. Training materials.

Ten nonwords were created for this study; all had three letters, three phonemes and a consonant-vowel-consonant structure. Only phonemes which are typically acquired by four years of age were used (Dodd et al., 2003), as children with Down syndrome often have phonological problems and exhibit more difficulties with later acquired sounds (Roberts et al., 2005). Additionally phonemes which could be depicted by multiple graphemes, e.g. /k/, were not used.

Flashcards of the spelling of the word were created for the orthography present training condition. For the orthography absent training condition, flashcards were created with the 'alien spelling' of the word, which consisted of three randomly selected Greek and Cyrillic letters. Previous studies (Ricketts et al., 2009; Rosenthal & Ehri, 2008) did not present a visual cue in their orthography absent conditions. However due to the younger chronological and mental ages of the typically developing children and the children with Down syndrome taking part in this study, it was considered important to control for the possibility that an extra cue (of any nature) may increase attention to the training situation.

Ten colour pictures were selected to fit with the theme of 'things found on an alien planet' and represented different semantic categories including food, animals, tools, transport, plants and housing. The pictures and the nonwords were randomly paired and then split into two groups (word group A and word group B) ensuring that nonwords with the same vowel pattern were in different groups (see Appendix 5 for a list of the nonwords, pictures and their pairings).

3.2.2.5. Learning procedure.

The children were introduced to the learning procedure by being told they were going to learn about an alien planet. They were told they would see pictures of things from the alien planet and learn what they were called. Each child was taught one word group, i.e. five nonwords, with the orthography present and the other word group with the orthography absent. The word group allocated to each condition was counter-balanced across participants. The two training conditions took place on different days, the order of which was also counterbalanced.

There were three repetition and three production trials during the learning procedure. All five words were trained within one trial and were presented in a fixed random order. Repetition and production trials were alternated. For each nonword, in the repetition trials children heard the word once, along with seeing the corresponding picture and a flashcard of the real spelling or the alien spelling. The children were either told “this is how we spell it” or “this is how they spell it on the alien planet”, and the flashcard and picture remained present for the duration of the trial. The children repeated the word, received corrective feedback and heard the word again. Most of the children in this study were able to read, therefore it was possible that they would be able to read the flashcards of the nonwords’ spellings. To attempt to equate stimulus exposure, the word was spoken by the experimenter one extra time in each trial in the orthography absent condition.

In the production trials, children were shown a picture (without the real or alien spelling) and asked if they could remember the name of the picture. They were given corrective feedback consisting of the spoken nonword and the appropriate flashcard. Attention was drawn to the flashcard in the same manner as above.

Children scored one point for each item they produced correctly in both the repetition and production trials. The production trials were the primary outcome measure from the learning procedure and consistent speech errors were taken into account when scoring children’s responses.

3.2.2.6. Experimental tasks.

Two post-tests were administered within the same session as the learning procedure but after a delay of 10-15 minutes. A picture naming post-test assessed how accurately children could produce the correct nonword for a given picture. The mispronunciation multiple-choice post-test was devised as a recognition task to assess the quality of children’s phonological representations of the taught nonwords without requiring production, similar to that of Jarrold et al. (2009).

3.2.2.6.1. Picture naming post-test.

In the picture naming post-test, children were shown the alien pictures they had learnt during that session and asked if they could remember their names. The pictures were presented individually in a fixed random order, and no flashcards were shown. Children scored one point for every item they produced correctly, and consistent speech errors were taken into account when scoring this test.

3.2.2.6.2. Mispronunciation multiple-choice post-test.

The mispronunciation multiple-choice post-test was a computerised task, in which the picture of an alien object taught that day appeared on the computer screen and pictures of three children were shown sequentially, each accompanied by a nonword, which was either the target nonword or a distracter nonword. The pictures of the three children were then shown simultaneously and the child was asked who had said the right name for the alien picture.

For each target word, two distracters were devised. For one distracter, the initial phoneme was changed by either place of articulation or voicing, and the final phoneme was changed in the same way for the second distracter, for example the distracters for target nonword 'tid' were 'pid' and 'tib' (see Appendix 5 for the distracters for all target nonwords). The target nonword and distracters were rotated across presentation position, i.e. first, second or third.

Prior to the experimental trials, two practice trials with real words and pictures were administered. For this task, and the following computerised tasks, all the target nonwords and the distracter words were recorded in a soundproof room and the tasks were presented on a laptop computer using e-Prime version 1.0 (Schneider, Eschman, & Zuccolotto, 2002).

3.2.2.6.3. Speech discrimination.

The distracters in the mispronunciation multiple-choice post-test were devised to be phonetically similar to the taught nonwords. However as children with Down syndrome are often reported to have hearing impairments (Shott et al., 2001), it was important to ensure that all participants could detect the difference between the correct phonemes and the substitutions used in the mispronunciation post-test. There were 11 such phoneme pairs, thus 11 pairs of nonwords four or five letters long were devised which differed only on these phonemes (e.g. brut and brup). A further 11 nonwords also with four or five letters were devised to act as filler items. Children listened to these nonword pairs on a laptop computer and had to decide whether they sounded the same or different. There were two practice trials with real words to illustrate the task requirements. There were two further practice trials with nonwords, before the 22 experimental trials were presented in a random fixed order.

3.2.2.7. Procedure.

Children took part in two sessions, each of which lasted 20-30 minutes. All sessions took place on an individual basis in a quiet area of the school. In the first session tasks were administered in the following order: vocabulary training, matrices, picture naming, single word reading, alliteration matching, picture naming post-test and the mispronunciation multiple-choice post-test. The second session followed the same format except the background measures administered were: speech discrimination, sound deletion and word recall.

3.2.3. Results

Raw scores and an alpha level of .05 were used for all analyses and data were complete for all measures.

3.2.3.1. Performance on background measures.

The performance of the children on the background cognitive measures is shown in Table 17. With the exception of alliteration matching, on which there are clear ceiling effects, there is a range of scores on all tasks suggesting the background tasks are appropriate for the target ability range. On the speech discrimination task, the children performed very accurately suggesting they had no difficulties discriminating between similar sounding phonemes.

3.2.3.2. Vocabulary learning.

The mean scores for the repetition trials can be seen in Table 18 and as would be expected children scored very highly on the repetition trials. This means that errors on production trials and the post-tests were not due to being unable to produce the nonwords. The mean scores for the children's levels of performance on the production trials during learning are shown in Figure 7. The scores for the production trials were low and children were at floor on the first trial. However there was some evidence of better performance in the orthography present condition, particularly on the last production trial.

A 2x3 ANOVA was conducted with condition (orthography absent vs. orthography present) and trials (1-3) as within-subject factors. There was a significant main effect of trial, $F(2,30)=11.09$, $p<.001$, $\eta_p^2 = .43$, indicating there was improvement during the learning

Table 17.

Performance on the background tasks for the typically developing children in Study 2a

	Mean score (standard deviation)	**Range**
Matrices (max. 29)	16.75 (5.50)	6-25
Single word reading (max. 60)	17.44 (11.32)	0-36
Alliteration matching (max. 10)	9.25 (0.68)	8-10
Sound deletion (max. 12)	8.00 (2.37)	4-10
Word recall (max. 42)	15.88 (2.09)	12-19
Picture naming (max. 30)	24.31 (2.80)	21-30
Speech discrimination target items (max. 11)	9.19 (2.71)	1-11

Table 18.

Results from the training and post-tests for the typically developing children in Study 2a

	Mean score (standard deviation)	
	Orthography absent	Orthography present
Repetition trials (max. 15)	14.81 (0.40)	14.75 (0.58)
Picture naming post-test (max.5)	0.56 (0.73)	0.94 (0.93)
Mispronunciation multiple choice post-test (max.5)	3.81 (1.17)	4.00 (1.03)

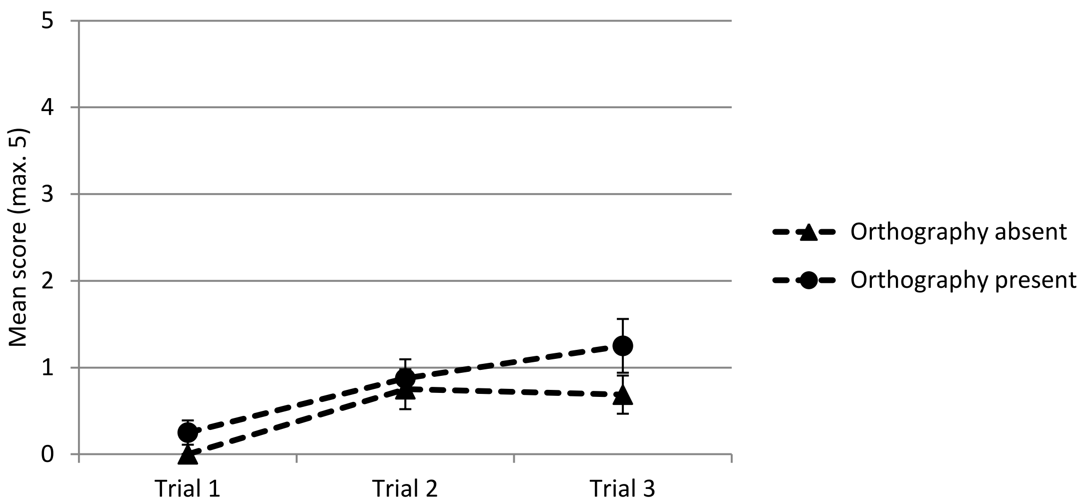


Figure 7. The mean scores for the production trials during vocabulary learning for the typically developing children in Study 2a with standard error bars

procedure. However there was no significant main effect of orthography or interaction between trial and orthography.

3.2.3.3. Post-tests.

The results for the picture naming and mispronunciation multiple-choice post-test are shown in Table 18. On the picture naming post-test the mean score was almost double in the orthography present condition than the orthography absent condition, although levels of performance were very low in both conditions. On the mispronunciation multiple-choice post-test performance was generally quite accurate, well above chance levels (i.e. 1.67). The scores across the two conditions were similar. Dependent t-tests showed that there were no differences between the two conditions for the picture naming and multiple choice mispronounciation post-tests.

3.2.4. Discussion

Study 2a was designed to trial a training procedure for teaching new spoken words with and without their orthography present with typically developing children before testing the children with Down syndrome. No significant differences were found between words taught with and without orthography during learning, on a picture naming post-test and a mispronunciation post-test.

Previous studies have found a beneficial effect from providing orthography during spoken word learning and on post-tests in typically developing children (Ricketts et al., 2009; Rosenthal & Ehri, 2008). Therefore the null results in this study were unexpected. There was a trend in the production trials, particularly towards the end of learning procedure, and on the picture naming post-test for scores to be higher in the orthography present condition. However this difference between the two conditions was not significant and scores were generally very low. In contrast, children were generally quite accurate on the mispronunciation multiple-choice post-test and there was no effect of orthography.

The low scores on the production trials and picture naming post-test suggest that children found it very difficult to learn the names of the novel pictures. If levels of learning were improved, this may result in a greater difference between the two conditions.

3.3. Study 2b

3.3.1. Overview

Study 2b was designed to address the limitations of Study 2a. Due to the difficulty that children experienced learning the nonwords, the number of trials was doubled with the aim of providing children with more practise and exposure to the phonology of the nonword and its picture pairing. Hence the general design of the training procedure stayed the same, although a second post-test was also added. The study only included typically developing children as it aimed to test this modified format of training before testing the children with Down syndrome.

3.3.2. Method

3.3.2.1 Design.

The same training format as Study 2a was followed. Ten spoken nonwords were paired with novel pictures and taught either with or without their orthography present. A mispronunciation forced-choice post-test was added.

3.3.2.2. Participants.

Six children (four males) from the same Year 2 classrooms as Study 2a took part in this second study and their mean age was 6;09. Children who had been identified with special educational needs were excluded. Consent was gained from both the headteacher of the school and parents.

3.3.2.3. Assessment battery.

The same assessment battery and training materials were used as Study 2a.

3.3.2.4. Learning procedure.

To increase learning levels, the number of trials was doubled to include six repetition and six production trials for each condition. The same procedure was followed as Study 2a.

3.3.2.5. Experimental tasks.

The picture naming and mispronunciation multiple-choice tasks from Study 2a were included. A mispronunciation forced-choice post-test was also added, to test how well children could identify whether they had heard the target nonword or a distracter when only presented with one option.

3.3.2.5.1. Mispronunciation forced-choice post-test.

Pictures of the alien object accompanied by a spoken nonword were shown on a computer screen individually. The nonword presented was a distracter for five of the items, and the correct answer for the remaining five (see Appendix 5 for items). Children had to decide whether the nonword they heard was the correct or incorrect name for the picture. Distracters were devised in the same way as for the mispronunciation multiple-choice post-test, but new distracters could only be constructed for five of the nonwords. Therefore the presentation of distracters and target nonwords could not be counterbalanced across all the items.

3.3.2.5.2. Speech discrimination task.

Due to the new distracters devised for the forced-choice mispronunciation task there was one additional phoneme pair to be discriminated between. Therefore this was added into the speech discrimination task, along with an extra filler pair resulting in 24 trials, 12 of which were target trials.

3.3.2.6. Procedure.

The same procedure was followed as in Experiment 2a, with the addition of the mispronunciation forced-choice task after the picture naming post-test and before the mispronunciation multiple-choice post-test.

3.3.3. Results

3.3.3.1. Performance on background tasks.

Children's scores on the background measures can be seen in Table 19. As in Study 2a there was a good range of scores on these tasks, with no floor or ceiling effects, with the exception of alliteration matching on which children generally scored very well. On the speech discrimination task, the children performed very accurately suggesting they were able to discriminate between the phoneme pairs utilised in the mispronunciation tasks.

3.3.3.2. Vocabulary learning.

The overall mean scores for the repetition trials can be seen in Table 20, and the scores for each production trial are shown in Figure 8. Children were very accurate on the repetition trials and the scores on the production trials were no longer at floor. There appeared to be an advantage for the orthography present condition in the production trials, and this was consistent throughout the learning procedure with scores for both conditions improving at a similar rate.

A 2x6 ANOVA with condition (orthography absent vs. orthography present) and trial (1-6) as within-subjects factors was conducted. The main effect of trial was significant, $F(5,25)=10.14$, $p<.001$, $\eta_p^2 = .67$, but the main effect of orthography and the interaction were not.

Table 19.

Performance on the background tasks for the typically developing children in Study 2b

	Mean score (standard deviation)	Range
Matrices (max. 29)	18.50 (3.94)	11-22
Single word reading (max. 60)	20.17 (9.02)	9-31
Alliteration matching (max. 10)	8.33 (1.97)	5-10
Sound deletion (max. 12)	7.33 (1.63)	5-9
Word recall (max. 42)	16.33 (2.73)	12-20
Picture naming (max. 30)	24.33 (1.51)	22-26
Speech discrimination target items (max. 12)	8.50 (3.99)	7-11

3.3.3.3. Post-tests.

The mean scores for the three post-tests can be seen in Table 20. Children were generally very accurate in both conditions on the two mispronunciation tasks. For the picture naming post-test, the mean scores were higher than in Study 2a but still quite low, and there appeared to be an advantage for the orthography present condition. Dependent t-tests were used to compare the two conditions on the picture naming, forced-choice mispronunciation and multiple-choice mispronunciation and no differences were significant.

Table 20.

Results from the vocabulary learning procedure and post-tests for the typically developing children in Study 2b

	Mean score (standard deviation)	
	Orthography absent	Orthography present
Repetition trials (max. 30)	27.17 (3.76)	28.33 (4.08)
Picture naming post-test (max.5)	1.33 (1.51)	2.17 (1.72)
Mispronunciation forced choice post-test (max.5)	4.50 (0.55)	4.50 (0.84)
Mispronunciation multiple choice post-test (max.5)	4.50 (0.84)	4.00 (1.26)

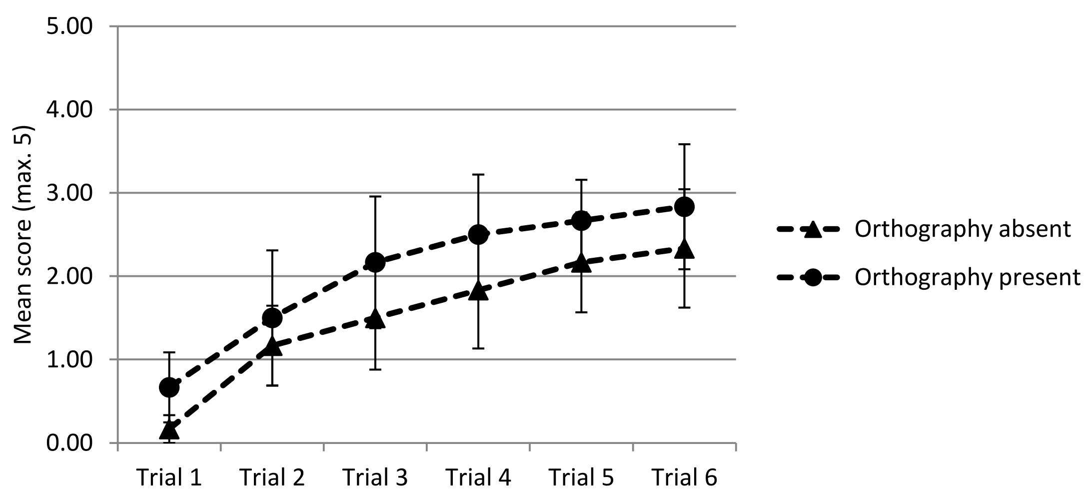


Figure 8. The mean scores for the production trials during learning for the typically developing children in Study 2b with standard error bars

3.3.4. Discussion

Study 2a aimed to devise a paradigm that would test whether orthographic support helps children when learning new spoken words. The results did not provide evidence for this hypothesis, although levels of learning were very low. Therefore the methodology was modified for Study 2b with the aim of improving children's learning. The number of trials was doubled and accordingly children's scores increased. A similar pattern of results was found for Study 2b as in Study 2a, where there appeared to be an advantage for the orthography present condition on the production trials and a picture naming post-test but this was not significant.

In contrast to the production trials and picture naming post-test, the children performed well on the two mispronunciation tasks but there were no differences between the two conditions. This may be because the typically developing children found these tasks too easy. However they may be more informative for children with Down syndrome for two possible reasons. Firstly there is some evidence that children with Down syndrome have difficulties with speech discrimination (Keller-Bell & Fox, 2007) and therefore might find this task more challenging, which in turn may lead to a greater opportunity for a benefit due to orthography. Secondly children with Down syndrome have articulation and phonological difficulties, and for this reason may find the picture naming post-test difficult. The mispronunciation tasks offer an opportunity to stringently assess the quality of the phonological representations of the new spoken words without requiring production.

Increasing the number of trials in Study 2b resulted in improved levels of learning on the production trials and picture naming post-test. However even at the end of the learning procedure children were making correct responses for approximately half of the items and even less than this for the picture naming post-test. During Studies 2a and 2b, children were frequently observed to be making two types of errors. There were matching errors where the children produced the correct phonological form of a taught nonword but in response to the incorrect picture. There were also mispronunciation errors where children were clearly attempting to say the correct nonword for the picture but made phonological errors. In previous research children have also been reported to make similar errors; McDuffie et al. (2007) identified difficulties with mapping the novel word to the correct stimulus and others have reported difficulties in producing the correct phonological form (Ehri & Wilce, 1979; Ricketts et al., 2009). Therefore the final training format was adapted to address these errors and thus aimed to increase learning further.

3.4. Study 2c

3.4.1. Overview

In Studies 2a and 2b, there was some evidence of an advantage for learning spoken words when the orthographic form was present. Although children's performance increased in Study 2b, learning still remained far from complete. Therefore the format of training was changed, with the addition of a trial which would enforce the links between the novel picture and nonword. It was hoped that this would improve learning by reducing matching errors. Activities were also added which explicitly segmented the target nonwords into their constituent phonemes, with the aim of improving children's phonological representations of the nonwords.

3.4.2. Method

3.4.2.1. Design.

Children with Down syndrome and typically developing children of a similar reading age participated in this final version of Study 2. The same ten nonwords as in Studies 2a and 2b were paired with pictures and taught with or without their orthography and all three post-tests from Study 2b were administered.

3.4.2.2. Participants.

3.4.2.2.1. Children with Down syndrome.

Sixteen children with Down syndrome were recruited from the larger sample of children taking part in Study 1. Information about the present study was sent to parents of children who were able to read at least one word and had speech which was largely intelligible. All parents contacted returned a consent form to allow their child to take part. An additional child who was not taking part in the longitudinal study also participated, resulting in a total sample of 17 children with Down syndrome (five males), who ranged in age from 7-16 years, and had a mean age of 12;09 (standard deviation of 2;10). According to parental questionnaires, 16 children had Trisomy 21 and one parent did not know what form of Down syndrome their child had. Seven children attended mainstream primary schools, seven children attended mainstream secondary schools and three children attended special secondary schools.

3.4.2.2.2. Typically developing children.

Twenty-seven typically developing children (11 males) were recruited from three primary schools from Reception, Year 1 and Year 2 classes. They were aged 5-7 years, with a mean age of 6;04 (standard deviation of eight months). Children who had been identified with special educational needs were excluded. Consent was gained from the headteachers of the schools and from parents.

The children were unselected but the year groups were chosen to correspond to the reading age range of the children. A larger normative sample that are not explicitly matched to the atypical group is an approach advocated by Jarrold and Brock (2004) to avoid issues such as generalisability and interpretation that can occur when groups are matched on a specific variable.

3.4.2.3. Assessment battery

The background measures of matrices, single word reading, picture naming, alliteration matching, sound deletion and word recall were administered as in Study 2a and 2b. In this final version of Study 2, there were differences in the nonverbal ability and reading tasks administered, as detailed below.

3.4.2.3.1. Nonverbal ability

The matrices subtest from the WPPSI-III^{UK} is normed for children aged up to 7;03. However because the testing of some of the typically developing group in the present study took place towards the end of the academic year, some of the children in Year 2 were older than this. Hence the WASI was administered to these children. The procedure for which version of the task to administer to the children with Down syndrome was the same as Study 1 (see section 2.2.3.1).

3.4.2.3.2. Reading accuracy.

Due to the inclusion of younger children in the final typically developing sample than in Studies 2a and 2b, a simpler task of word reading, the early word reading test from the YARC Early Reading battery was administered to all children. Details of this task can be found in section 2.2.3.3.2.

3.4.2.4. Learning procedure.

The vocabulary learning procedure was divided into four cycles. Each cycle included three different trials, and the cycles were designed to increase in difficulty. There were two differences from the format of training in Studies 2a and 2b: a phonological consolidation exercise and a matching game.

The first trial consisted of repetition and phonological consolidation. The repetition activity was identical to the previous training formats; children heard the nonword, saw the picture, repeated the nonword and received corrective feedback. The phonological consolidation aspect differed slightly in each cycle of training. In Cycle 1 children heard the word sounded out, repeated it and heard the initial sound isolated. In Cycle 2 children had to produce the initial sound independently, they were given corrective feedback and heard the word sounded out. Cycles 3 and 4 followed the same format, except the focus was on the final, rather than initial, sound. The real spelling or alien spelling flashcard was present throughout.

The second trial was a matching game presented on a laptop computer in a Microsoft Office PowerPoint presentation. The children heard the nonword and had to identify the corresponding picture shown on the computer screen. Children then received corrective feedback in which they also heard the word again. The real or alien spelling was present on the computer screen throughout. The cycles increased in difficulty by including one, two, three or four of the other pictures being trained in that session as distracters.

The third trial was a production trial and this was the same as in Studies 2a and 2b. Children were presented with a picture and asked to name it. They were given corrective feedback, heard the correct word and saw the relevant flashcard.

As in the previous studies, the children heard the word an extra time in each trial during the orthography absent training to equate for stimulus exposure and consistent speech errors were taken into account when scoring responses.

3.4.2.5. Experimental tasks.

The experimental tasks from Study 2b were also included here: the picture naming post-test, mispronunciation forced-choice post-test, mispronunciation multiple-choice post-test and speech discrimination task.

3.4.2.6. Procedure.

3.4.2.6.1. Typically developing children.

The training sessions lasted 30-40 minutes each. The sessions followed the same format as the previous experiments, and early word reading was administered in the first session before single word reading. Therefore where possible the first session included, in order: vocabulary learning, matrices, picture naming, early word reading, single word reading, alliteration matching, alien picture naming post-test, the mispronunciation forced-choice post-test and the mispronunciation multiple-choice post-test. Where possible the second session included, in order: vocabulary learning, speech discrimination, word recall, sound deletion, alien picture naming post-test, the mispronunciation forced-choice post-test and the mispronunciation multiple-choice post-test. Testing took place in a quiet space within the school and children were seen individually. The delay between sessions varied between two and 20 days, with a mean of 5.70 days.

3.4.2.6.2. Children with Down syndrome.

Sixteen of the children with Down syndrome were also taking part in Study 1, and the test battery for this included matrices, picture naming, early word reading, single word reading and alliteration matching. For 12 of these children the vocabulary training study took place at the same time as testing for Time 2 of the longitudinal study. For the remaining four children, the vocabulary training and measures not included in Study 1's test battery were administered 2-4 months later. Where possible the tasks were administered in the same order as the typically developing group. For the child who was not taking part in the longitudinal study, the training sessions followed the same format as for the typically developing group. The delay between sessions was typically longer for the children with Down syndrome due to the logistics of testing in different schools. The mean delay was 13.71 days, and ranged from four to 37 days.

3.4.3. Results

Raw scores were used for all analyses, except the matrices task. For this, different participants completed different versions according to their age or ability, and therefore age-equivalent scores are presented for this task.

Most of the data were complete, although there were a few exceptions. One child with Down syndrome refused to complete the post-tests, along with the speech discrimination and sound deletion tasks. Another child with Down syndrome did not complete the speech discrimination, word span and sound deletion tasks. One child from the typically developing group did not complete the speech discrimination task.

3.4.3.1. Performance on background measures.

To examine the distribution of scores within the two groups the Shapiro-Wilk test was used and histograms, along with measures of skew and kurtosis, were examined. For both groups, the matrices scores were positively skewed and the early word reading task was negatively skewed. The distribution for the picture naming task in the typically developing group was clustered and negatively skewed, as most children scored between 20 and 30. There were ceiling effects for the alliteration matching task in the typically developing group, with 15 of the children scoring maximum points. The typically developing group were very accurate on the speech discrimination task, leading to a negative skew.

If the distribution for a task deviated from normal for either or both groups then a Mann-Whitney U test was used to test the differences between the groups. If the distributions were normal in both groups, then an independent t-test was used. The mean scores, standard deviations and between-group tests are reported in Table 21. As would be expected from previous literature, there were no differences between the groups on the two reading accuracy tasks but the typically developing group performed significantly better on all other tasks.

3.4.3.2. Vocabulary learning.

The mean scores for the children's performance on the repetition trials are shown in Table 22 and it can be seen that both groups of children performed very accurately on the repetition trials. The scores on each of the four production trials are shown in Figure 9, and there appeared to be higher scores in the orthography present condition. Learning was evident across the production trials in both conditions but there was some evidence for an increasing advantage for orthographic support. The two groups performed very similarly throughout the learning procedure. The maximum score for the production trials was 20. The typically developing children read a mean of 5.26 words correctly in the orthography absent condition and 9.35 in the orthography present condition overall. The group of children with Down syndrome read a mean of 5.78 words correctly in the orthography absent condition and 8.85 in the orthography present condition. It should be noted therefore that learning was not complete in either group.

A mixed 2x4x2 ANOVA was performed for the production trials, with condition (orthography absent vs. orthography present) and cycle (1-4) as within-participants variables and group (Down syndrome vs. typically developing) as a between-participants variable. Mauchly's test indicated that the assumption of sphericity had been violated for the main effect of cycle, $\chi^2(5) = 13.58$, $p=.019$, and therefore degrees of freedom for this variable were corrected using Greenhouse-Geisser estimates of sphericity ($\varepsilon=.82$). The main effect of condition was significant, $F(1, 42)=23.52$, $p<.001$, $\eta_p^2 = .36$, due to higher scores in the orthography present condition. There was also a main effect of cycle, $F(2.45, 104.76)=60.80$, $p<.001$, $\eta_p^2 = .59$, due to scores increasing during the learning procedure. There was no main effect of group, and no significant interactions involving group. There was a significant interaction between condition and cycle, $F(3,126)=7.74$, $p<.001$, $\eta_p^2=.16$. As can be seen from Figure 9 this is because there was a greater improvement for the orthography present condition than the orthography absent condition.

Table 21.

Scores of the children with Down syndrome and typically developing children on all background measures (Study 2c)

	Scores of the children with Down syndrome		Scores of the typically developing children		
	Mean score (standard deviation)	Range	Mean score (standard deviation)	Range	Between-group differences
Matrices (age-equivalent score)	5;00 (1;04)	4;00-8;06	6;07 (2;04)	4;00-14;06	U=352.00, *p*=.003
Early word reading (max. 30)	24.71 (7.28)	5-30	23.81 (9.68)	0-30	U=247.50, *p*=.639
Single word reading (max. 60)	23.24 (12.49)	3-40	24.15 (14.55)	0-47	t(42)=-.21, *p*=.832
Alliteration matching (max. 10)	6.76 (1.60)	5-9	8.78 (1.95)	3-10	U=379.00, *p*<.001
Sound deletion (max. 12)	4.93 (3.03)	0-9	8.52 (3.36)	2-12	t(40)=-3.43, *p*=.001
Word recall (max. 42)	13.25 (3.96)	6-23	17.70 (4.43)	9-26	t(41)=-3.31, *p*=.002
Picture naming (max. 30)	20.59 (4.20)	10-25	24.37 (3.92)	10-30	U=349.00, *p*=.004
Speech discrimination target items (max. 12)	6.40 (3.07)	3-12	9.92 (2.33)	4-12	U=312.50, *p*=.001

Table 22.

Mean scores (and standard deviations) for the children with Down syndrome and the typically developing children on vocabulary training and post-tests (Study 2c)

	Children with Down syndrome		Typically developing children	
	Orthography absent	Orthography present	Orthography absent	Orthography present
Total repetition trials (max.20)	19.00 (1.87)	19.59 (0.87)	19.96 (0.19)	19.70 (0.78)
Picture naming post-test (max. 5)	1.44 (1.09)	2.75 (1.65)	1.59 (1.74)	2.81 (1.78)
Forced choice mispronunciation post-test (max.5)	2.81 (0.75)	3.06 (0.68)	4.07 (1.04)	4.07 (1.11)
Multiple choice mispronunciation post-test (max.5)	3.31 (1.35)	3.19 (1.42)	4.04 (1.26)	4.00 (1.33)

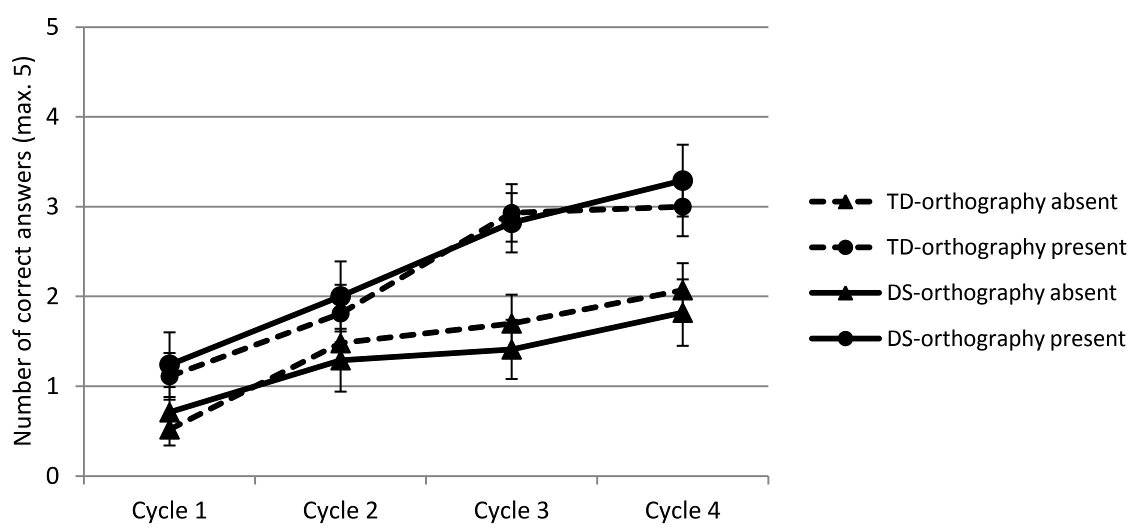


Figure 9. The mean scores for the production trials during vocabulary learning for the typically developing children and children with Down syndrome with standard error bars (Study 2c)

3.4.3.3. Post-tests.

The mean scores for performance on the picture naming, mispronunciation forced-choice and mispronunciation multiple-choice post-tests are shown in Table 22. For the picture naming post-test the two groups performed similarly. More items were answered correctly in the orthography present condition although scores were not particularly high. A different pattern emerged for the mispronunciation tasks. The typically developing group scored more highly than the children with Down syndrome on both tasks, and there did not appear to be a benefit from the presence of orthography. 2x2 mixed ANOVAs, with condition (orthography absent vs. orthography present) as a within-subjects variable and group (Down syndrome vs. typically developing) as a between-subjects variable, were conducted for each post-test.

For the picture naming post-test there was a main effect of orthography, $F(1, 41)=36.70$, $p<.001$, $\eta_p^2 = .47$, due to better performance in the orthography present condition. The main effect of group and the interaction between group and orthography were not significant.

For the mispronunciation forced-choice and multiple-choice tasks, there were main effects of group, $F(1, 41)=22.85$, $p<.001$, $\eta_p^2 = .36$, and $F(1, 41)=5.32$, $p=.026$, $\eta_p^2 = .16$ respectively. In both cases this was due to the lower performance of the children with Down syndrome. The main effect of orthography and the interaction between orthography and group were not significant for either task.

3.4.3.4. Error analysis

Learning a new spoken word involves a number of different processes. Firstly the phonology of the new word has to be learnt accurately and precisely, the referent or semantics (in this case, the picture) also has to be learnt, and the mapping between the word and referent has to be established. The errors children made during the learning procedure could indicate which aspect of the learning process they may have difficulties in. Although the children with Down syndrome and typically developing children answered the same number of items correctly, they may have made different errors suggesting underlying differences.

Errors were classified into several categories: don't know/no response, phonological, mismatch or unrelated. The phonological errors were further broken down into which phonemes were produced correctly. A mismatch error occurred when a different taught nonword was produced. This would indicate problems with establishing links between the nonwords and the specific pictures they were paired with. If the link had been established but the phonological representation of the nonword was poor then a phonological error would arise.

It could be hypothesised that as children with Down syndrome have weaknesses in phonology, then this could manifest itself in a greater preponderance of phonological errors. However when the number of errors in each category were analysed, no significant differences were found between the two groups on any error type. This suggests that not only are children with Down syndrome and typically developing children reaching the same endpoint of learning, they are behaving similarly during the learning process and on the different aspects of learning a new spoken word.

3.4.3.5. Summary of word learning performance.

Children with Down syndrome performed equivalently to typically developing children when producing the names of novel pictures during training and after a 10-15 minute delay. Furthermore orthographic support improved learning for both groups to the same extent. However on mispronunciation post-tests, orthography was not found to affect performance and the typically developing group performed significantly better than the children with Down syndrome.

3.4.3.6. Correlations between the background measures and performance on the word learning task.

It was of interest to examine if any of the cognitive skills assessed were related to children's spoken word learning and if this differed in the two groups. Therefore correlations were conducted between the background measures and the production trials. The production trials in each condition were not strongly correlated with each other in both groups: r=.63** in the children with Down syndrome and r=.59** in the typically developing group. Therefore

Table 23.

Simple correlations between the background measures and production trials for the children with Down syndrome and typically developing children (Study 2c)

	Children with Down syndrome		Typically developing children	
	Orthography absent	Orthography present	Orthography absent	Orthography present
Age	-.03	-.02	.30	.44*
Matrices	-.40†	.11	.39*†	.50**
Reading accuracy	.17	.45	.63**	.66**
Alliteration matching	-.04	.26	.44*	.47*
Sound deletion	.17	.55*	.56**	.61**
Word recall	.35	.72**	.49**	.36
Picture naming	.19	.52*	.48*	.46*
Speech discrimination target items	.53*	.56*	.16	.41**

Note. †significant difference between the correlations in the two groups
*p<0.05 **p<0.01

separate correlations are reported for the orthography present and orthography absent production trials. As in Study 1, the different measures of reading accuracy, in this case single word reading and early word reading, were correlated strongly in the typically developing children and children with Down syndrome (r=.84** in both groups). Therefore the z-scores from these two measures were averaged to form a reading accuracy composite, which will be used in the following correlations.

The simple correlations between the background measures and the learning tasks can be seen in Table 23. For the children with Down syndrome, the speech discrimination task was significantly correlated with both learning conditions. However many of the other background tasks were significantly correlated with learning in the orthography present condition but not the orthography absent condition, with word recall being a particularly strong correlate. In the

Table 24.

Partial correlations controlling for age and matrices between the background measures and production trials for the children with Down syndrome and typically developing children (Study 2c)

	Children with Down syndrome		Typically developing children	
	Orthography absent	Orthography present	Orthography absent	Orthography present
Reading accuracy	.37	.44	.45*	.51*
Alliteration matching	.18	.24	.24	.30
Sound deletion	.38	.56*	.42*	.45*
Word recall	.44	.72**†	.20	.02†
Picture naming	.51[a]	.55*	.27	.23
Speech discrimination target items	.57*†	.58*	-.02†	.30

Note. [a] p=.054
†significant difference between the correlations in the two groups
*p<0.05 **p<0.01

typically developing group, most of the correlations between the background measures and the two learning conditions were moderate. There was a significant difference between the two groups, with matrices being more strongly correlated with orthography absent learning in the typically developing children.

Partial correlations were also conducted to control for age and nonverbal ability; these are shown in Table 24. Only reading accuracy and sound deletion remained moderately correlated with the learning measures in the typically developing group. For the children with Down syndrome, speech discrimination was significantly correlated with both learning conditions and word recall, picture naming and sound deletion were correlated significantly with the orthography present condition. The correlation between picture naming and the orthography absent condition was marginally significant. There were two instances of stronger correlations in the group of children with Down syndrome than in the typically developing group: speech discrimination with orthography absent learning and word recall with orthography present learning.

3.4.4. Discussion

The aim of Study 2c was to examine spoken word learning in children with Down syndrome and more specifically to test whether the presence of the orthographic form benefits learning of the phonological form. This was compared to a group of typically developing children of similar reading ability and it was expected that the typically developing children would show greater levels of word learning. This was the case on the two mispronunciation post-tests, but more importantly the two groups performed similarly during learning and on a picture naming post-test. It was predicted that both groups would show a benefit from orthography and this study aimed to explore if this would be to the same extent in both groups. It was found that orthographic support did lead to higher levels of performance during learning and on the picture naming post-test, and this benefit was similar in the two groups. However there was no effect of orthography on the mispronunciation post-tests.

The findings from this study suggest that children with Down syndrome do not have a relative impairment on tasks that require accurate production of novel words, in support of Mosse and Jarrold (2011). However, it should be emphasised that learning was only assessed immediately after training and not in the longer term. The performance of the children with Down syndrome was significantly poorer than the typically developing group on a measure of existing vocabulary knowledge. Therefore there appears to be a disparity between children with Down syndrome's ability to acquire, store and retrieve a word on the same day and the storage and retrieval of words over prolonged periods of time.

Both groups of children benefited from orthography during the learning procedure and on the picture naming post-test. This effect is in line with previous research using similar paradigms with typically developing children (Ricketts et al., 2009; Rosenthal & Ehri, 2008), and extends the findings to children with Down syndrome. The present study utilised a control condition that consisted of three non-orthographic symbols, which highlights that the benefit from orthography is because it provides meaningful information about the new word, rather than just presenting extra information or acting as a general visual cue. It is argued that orthography provides children with a means of confirming the phonology of the new word using grapheme-phoneme correspondences resulting in a more accurate phonological representation. Furthermore, in line with the lexical quality hypothesis (Perfetti & Hart, 2002), orthography provides another representation of the new word form in memory that aids retrieval. Treiman and Bourassa (2000) have also found that children were more accurate when they spelt words or nonwords on paper rather than orally, and even their incorrect attempts were more likely to resemble the target phonological structure. They suggested that if children do not have an existing representation of a word's spelling then they need to segment the word into its constituent phonemes and this is done most easily when it is in a visible and permanent form.

A beneficial effect of reading on spoken language has been reported for children with Down syndrome but this is the first study to examine whether this is greater in children with Down syndrome than in typically developing children. Due to strengths in visual memory and word recognition and weaknesses in expressive language, hearing and verbal memory, it could be expected that children with Down syndrome would benefit more from orthography (Buckley & Bird, 1993). However the two groups were facilitated equally by the provision of orthography, presumably because they had similar reading skills and therefore were able to access the orthography to the same extent.

The mispronunciation post-tests were designed to assess children's phonological representations without requiring expressive language. In contrast to the production trials and picture naming post-test, typically developing children performed significantly better than the children with Down syndrome and there was no effect of orthography. The relatively poorer performance of the group of children with Down syndrome is in line with Jarrold et al. (2009) but in contrast to Mosse and Jarrold's replication (2011). This group difference may be due to task demands which are unrelated to children's phonological representation of the novel word, for example difficulties with understanding instructions or remembering which position the correct nonword had been presented. Furthermore, children with Down syndrome performed significantly poorer on a speech discrimination task than the typically developing children, which tested their ability to discriminate between the distracter and target items used in the mispronunciation tasks. Considering the effects of orthography, there was no difference between the two training conditions on the mispronunciation post-tests. This may be because even in the orthography absent condition children's phonological representations were accurate enough to complete this task, and therefore there was no additional advantage provided by orthography. It could also be due to methodological reasons; the maximum score on these tasks was five and the choice of answers was either three or two, hence by chance children would have scored 1.67 or 2.50, respectively. This therefore reduces the potential for a benefit from orthography.

The relationship between the cognitive tasks and spoken word learning was examined. For children with Down syndrome, reading, vocabulary knowledge, verbal short-term memory, sound deletion and speech discrimination were correlated moderately to strongly with learning. The correlation with memory was particularly strong and supports Jarrold et al. (2009) who found that phonological learning was best predicted by verbal short-term memory in children with Down syndrome. For typically developing children, reading and sound deletion were the measures most highly correlated with learning. This supports previous research in typically developing children, which has also found that reading and phonological awareness correlated with spoken word learning (Ehri & Wilce, 1979; Hu, 2008; Rosenthal & Ehri, 2008).

This study aimed to investigate whether reading can aid oral language learning in children with Down syndrome by presenting the written form of a word whilst teaching its spoken form. There is also some evidence that when learning new sentences, children with Down syndrome are more accurate when they have previously seen the written form of the sentence (Buckley, 1993). Both morphology and syntax are particular weaknesses for children with Down syndrome (Laws & Bishop, 2003) and therefore it is possible that orthography may particularly benefit these skills in children with Down syndrome compared to typically developing children. This could be tested experimentally using a similar paradigm to the present experiment, for example correct sentence structures could be taught using a short video or series of pictures with an accompanying spoken and written sentence. Pictures could also be used to teach new morphemes with the target morphemes being highlighted in the accompanying written word.

The focus of this spoken word learning study was on phonology but in everyday life there is also a stronger semantic component. When presented with the orthography of a spoken word, a benefit on semantic learning has also been found in typically developing adults and children (Nelson, Balass, & Perfetti, 2005; Ricketts et al., 2009; Rosenthal & Ehri, 2008). It would be interesting to explore this in children with Down syndrome and investigate whether similar beneficial effects of orthography would be found for semantics as well as phonology. The format of training used in this study could be easily adapted to include deeper semantic information such as definitions or story contexts.

3.4.4.1. Summary and conclusions.

In summary, this study has demonstrated that children with Down syndrome are able to learn the phonological form of new words to the same level as typically developing children matched on reading, and that they benefit from orthography to the same extent as typically developing children. A practical application of this work is that children would benefit from being shown a flashcard of the written form of a word when learning its spoken form. This lends empirical support to current practice recommended for children with Down syndrome (Bird, Alton, & Mackinnon, 2000) but also highlights a similar potential benefit for typically developing children. Further research should investigate whether this orthographic advantage extends to semantic learning of new words and other domains of spoken language such as grammar.

Chapter Four

Orthographic Learning in Children with Down Syndrome and the Role of Phonological Pre-Training

4.1. Introduction

4.1.1. Overview

In typical development children become sensitive to the relationship between orthography and phonology at a young age and are able to use this information to decode unknown words. Decoding is a necessary skill to read unfamiliar words but it is effortful and can be unsuccessful for irregular words. Skilled readers do not need to decode familiar words; they are able to retrieve word pronunciations from memory using their orthographic representation of the word (Ehri, 2005a). This successful orthographic learning is argued to arise due to the connections between the orthography and phonology of a word, which are formed by the application of the alphabetic principle (Ehri, 2005a).

This chapter will first discuss existing research that has examined orthographic learning, with a particular focus on children with reading difficulties and the role of phonological skills in typically developing children. No existing studies have investigated orthographic learning in children with Down syndrome. However such studies offer a way of examining the relationship between orthographic learning and phonology in children with Down syndrome and whether this differs from typical development, as with the relationship between acquired reading skill and phonological awareness. Specifically the effect of phonological pre-training can be compared in the two populations to assess whether this benefits orthographic learning and to what extent.

4.1.2. Self-teaching

Share's self-teaching hypothesis is a theory about how children learn to read new words (Jorm & Share, 1983; Share, 1995). It is argued that when children encounter an unknown word, they independently convert the letters into sounds, a process termed phonological recoding. Multiple successful encounters result in the formation of an orthographic representation for that particular word. Share proposed that phonological recoding begins on an individual letter basis and progresses to a more lexicalised process utilising analogies and morphological units. There is a clear role for phonological skills in this process and orthographic skills are argued to have a secondary influence.

The self-teaching paradigm developed to assess this theory involves children reading aloud stories that contain target nonwords. This is done independently and no corrective feedback is given. Post-tests are then given to assess orthographic learning, either immediately or after a delay. Typically there are three tasks administered: orthographic choice, naming, and spelling. The orthographic choice task requires children to identify the target nonword when it is presented with a pseudohomophone (a nonword which shares the phonology of the target word but differs in its orthography), for example if the target word is 'yait' then the pseudohomophone could be 'yate'. Pseudohomophones are also presented, along with the target nonwords, in naming tasks. In spelling tasks, children's responses can be classified as correct or as a pseudohomophone. If children perform well on these tasks, and show an advantage for the taught spelling over the pseudohomophone, then this would suggest that they have learnt the specific spelling pattern associated with the target nonword rather than just the phonology. Indeed, English-speaking and Hebrew-speaking children aged seven to eight have been found to choose the target nonword more than a pseudohomophone in an orthographic choice post-test, name the target nonword faster than a pseudohomophone and produce the correct spelling more often than a pseudohomophonic spelling (Cunningham, Perry, Stanovich, & Share, 2002; Share, 1999).

A key premise in the self-teaching hypothesis is the causal role of phonological skills in forming orthographic representations. A prediction from this would be that when phonological recoding is impeded, orthographic learning should decrease. To test this, Share (1999) instructed children to repeatedly articulate an unrelated word whilst completing a lexical decision task with the target nonwords. This aimed to reduce opportunities for phonological recoding and indeed orthographic learning on all three of the standard post-tests was reduced compared to the classic self-teaching paradigm.

Share's (1999) comparison was between different children and across different tasks, and these limitations were addressed by Kyte and Johnson (2006). English-speaking children aged 10 years completed two lexical decision tasks, one where they read the nonword aloud and a second where they had to repeat the syllable 'la'. The children showed better performance for nonwords which had been read aloud on all three self-teaching post-tests, although the advantage for naming was relatively modest.

The findings of Share (1999) and Kyte and Johnson (2006) provide evidence for a role of phonological recoding in creating and storing orthographic representations. Notably in both these studies, orthographic learning still occurred under the concurrent articulation conditions. This may indicate that other factors, such as visual skills, also play a role in orthographic learning. It could also be that phonological recoding was not entirely suppressed. It is perhaps most likely that both of these processes played a role.

The number of exposures to the target nonword can be manipulated in the self-teaching paradigm to explore how many times children need to see a word to create and store an orthographic representation. Share (2004) compared the effect of one, two and four exposures of the target nonwords with children aged 8-9 years. He found that orthographic learning occurred in all three conditions and there were no differences between them. For the same age group of children Bowey and Muller (2005) found an advantage for words presented eight times compared to four times on an orthographic choice post-test. The discrepancy between the findings could be due to the relatively lower power in Share's study as there were more comparisons (Bowey & Muller, 2005), or the difference between the number of exposures examined.

The durability of the newly created orthographic representations has been examined by varying the amount of time between story-reading and the post-tests. Share (2004) found that performance on all three standard post-tests was equivalent after a three day, one week and one month delay. Bowey and Muller (2005) included a delayed post-test after six days, as well as an immediate post-test using an orthographic choice task. There were higher levels of learning on the immediate post-test than the delayed post-test. Bowey and Muller suggest this is because greatest degrees of learning occur immediately, resulting in better performance on an immediate post-test, and then this declines very quickly and reaches a stable level, which is reflected by the consistent performance across the delayed post-tests as found by Share.

4.1.3. Orthographic Learning in Single Word Contexts

Orthographic learning has also been studied using alternative paradigms to self-teaching. Typically this takes the form of training studies where children learn to read new words in isolation receiving corrective feedback. Reitsma (1983) carried out one such study with 14 children aged 7-8 years. Children heard and repeated nonwords and then were asked to read aloud the same nonwords four or eight times. Three days later children undertook a naming post-test. Children who received eight exposures during orthographic learning read target nonwords faster than pseudohomophones, whereas there was no such difference for the group of children who read the nonwords four times. This effect supports the findings of Bowey and Muller (2005) suggesting that more exposures to new words result in better orthographic representations.

The pronunciation of words can be either consistent or inconsistent with the orthography, and this variable was manipulated in Ricketts, Bishop and Nation (2008). For the consistent items, the vowel pattern was trained to be pronounced in the most common manner and for the inconsistent items the trained pronunciation was the same as only one or two words with the same vowel pattern, for example the word ‘mouge’ was trained with a pronunciation which either rhymed with ‘gouge’ (consistent) or ‘rouge’ (inconsistent). Children aged 9-10 years heard, saw and repeated the nonword. They were then trained to read the nonwords presented in isolation before reading the nonwords in stories with corrective feedback. During the training and on spelling and orthographic choice post-tests, children performed significantly better on nonwords with a consistent pronunciation. This advantage for consistent pronunciations has also been found with adults (Burt & Blackwell, 2008) and suggests that when phonology maps on to orthography in an easily decodable way, reading attempts tend to be more successful and hence a correct orthographic representation is more likely to be formed.

4.1.4. The Effect of Pre-Existing Phonological Representations on Orthographic Learning

Outside of experimental contexts, when children encounter unknown words in print they may already be familiar with them in the oral domain. A pre-existing phonological representation may make it easier for children to read new words and therefore to establish an orthographic representation. Indeed Share (2004) found that in a self-teaching task words chosen to be familiar in their spoken form but rare in their written form, were decoded more accurately than nonwords.

It is easier to control for pre-exposure to the spoken form of the words to be learned by teaching children to read nonwords. McKague et al. (2001) asked children aged 6-7 years to listen to and repeat nonwords before they attempted to read the nonwords. Children were significantly more accurate when reading pre-trained nonwords than control nonwords which had not been pre-trained. In a second experiment McKague et al. found that this advantage also existed when children merely listened to the nonword, rather than being required to repeat it, suggesting that even brief and shallow exposures to the phonology of the nonword provides benefits.

Duff and Hulme (in press) carried out a similar experiment with slightly younger children aged 5-6 years. During the phonological pre-training children heard the nonword, repeated it and had to discriminate between the target nonword and phonologically similar distracters. Pre-trained and control nonwords then underwent orthographic training where children read the nonwords individually and received corrective feedback. Six days later children attempted to read the nonwords again. Nonwords which had received phonological pre-training were read more accurately throughout training and in the post-test.

In summary, when children are familiar with the phonology of a nonword they are more likely to be able to read the nonwords. However the effect on orthographic learning post-tests, which assess the quality of the orthographic representation rather than children's decoding skill, has not been tested.

4.1.5. Orthographic Learning in Children with Reading Difficulties

The dominant theory of dyslexia places a causal role on phonological deficits (Hulme & Snowling, 2009). As orthographic learning is directly affected by phonological skills, it is of interest to examine orthographic learning in children with reading difficulties. Furthermore the question arises as to whether children with reading difficulties would perform equivalently on a dynamic orthographic learning task as typically developing children who are at the same level on a test of acquired reading skill. If children with reading difficulties acquire new orthographic representations at a slower rate, which is likely as they are often older than typically developing control groups, then they may be impaired on learning tasks.

According to Share (1995), children with poor phonological skills can still self-teach via a rudimentary mechanism using a basic level of phonological awareness, letter-sound knowledge and context to complete partial decoding attempts. However as self-teaching relies heavily on independent phonological recoding, it would be expected that individuals who have weak phonological skills may show impairments. Share and Shalev (2004) tested this hypothesis with four groups of children: children with dyslexia aged 9-12 years, 'garden variety' poor readers of the same age (also with low IQs), a chronological age matched control group and a younger control group aged 7-8 years who were matched on reading ability to the garden variety group only. Despite having equivalent or lower target decoding skill, both poor reader groups were able to show some evidence of orthographic learning whereas the younger control group did not, which the authors attribute to lower levels of print exposure and thus poorer orthographic processing skills. The poor reader groups performed more poorly than the age matched group on target decoding during story reading and post-tests of orthographic choice and naming. Importantly, when reading skill was controlled for, these differences between the groups no longer existed, indicating that lower levels of reading ability were the reason for the poor readers' difficulty.

Orthographic training studies in single word contexts have also included groups of children with reading difficulties. Reitsma (1983) trained children to read words which were likely to be familiar in the spoken form to children but unfamiliar in print. Thirteen children who had reading difficulties and a mean age of 9;08, and 18 typically developing children aged

6-7 years matched on reading ability read sentences containing the target words. In a naming post-test three days later, the typically developing group read the trained words faster than pseudohomophones, whereas there was no such difference for the group of children with reading difficulties. This suggests that the children with reading difficulties had not acquired a precise orthographic representation of the trained words. In contrast to Share and Shalev (2004) differences in orthographic learning are unlikely to be attributable to reading level as the groups were matched on this variable.

Ehri and Saltmarsh (1995) found differences between children with reading difficulties and typically developing children during orthographic learning as well as on post-tests. They taught simplified spellings of words (e.g. 'mesngr' for 'messenger') whose spoken forms should be familiar but written forms should be unfamiliar to 30 typically developing children aged 6-7 years and 15 children with reading difficulties aged 7-10 years. Children were shown the spellings, heard the word in a defining sentence and had to repeat it. They then attempted to read the nonwords up to 12 times receiving corrective feedback. When controlling for reading ability, children with reading difficulties required more trials to read the taught words correctly. In a naming post-test, only the typically developing children showed an advantage for the taught words compared to pseudohomophones. These results suggest that although children with reading difficulties showed measurable levels of orthographic learning, this was slower than their acquired reading level would suggest and they had weaker orthographic representations of the taught words. It is possible that the children in this study and Reitsma (1983) had phonological difficulties beyond their reading level and therefore may be less able to fully decode the new words. This would result in partial grapheme-phoneme associations being formed and thus weaker and incomplete orthographic representations.

The possible impact of different subtypes of reading difficulties and underlying differences in phonological awareness was examined by Bailey, Manis, Pedersen and Seidenberg (2004). Four groups of thirteen children took part: one group of children with phonological dyslexia, one group of children with surface dyslexia, one typically developing group matched on chronological age and one younger typically developing group matched on reading skill. Children with phonological dyslexia were classified as having difficulties with nonword reading, whereas children with surface dyslexia had difficulties with exception word reading. Children received phonological pre-training for either a regular or exception pronunciation and then attempted to read the nonwords independently. During orthographic learning both groups of children with dyslexia performed significantly poorer than the typically developing group of the same chronological age. The children with phonological dyslexia performed significantly worse on the regular words than their reading matched control group, and as such were the only group not to show an advantage for the regular words compared to the exception words. This suggests that children who have intact phonological skills but difficulties reading exception words show a delay in orthographic learning that is in line with their reading skill. Children with underlying phonological difficulties and weaknesses in decoding however, show deficits beyond their acquired reading level.

4.1.6. Summary

Phonological skills are necessary to successfully decode novel words and therefore to create accurate orthographic representations. Typically developing children are able to do this independently as shown by their success in self-teaching paradigms. Testament to the key role of phonology, nonwords which receive phonological pre-training are more successfully read although the effect on orthographic post-tests is unknown.

Children with poor reading skill have been found to have difficulties with orthographic learning. The studies reviewed above have mixed conclusions as to whether this is in line with or below their acquired reading skill. These differences may be due to different subtypes of reading difficulties as children with more severe phonological deficits have weaker orthographic learning beyond that expected from their reading level (Bailey et al., 2004). This suggests that they may acquire new orthographic representations at a slower rate than typically developing children at the same reading age. However it must be noted that there are other skills involved in orthographic learning such as orthographic processing skills, which may play a larger role in children with reading difficulties as they are older than reading matched control groups and therefore may have had greater levels of print exposure (Cunningham et al., 2002; Share & Shalev, 2004).

4.1.7. Introduction to Study 3

There are no experimental studies which have examined orthographic learning in children with Down syndrome. This population typically has relative strengths in word recognition compared to nonword reading and phonological skills. Therefore the processes underlying the acquisition of orthographic representations may be different to typically developing children.

Like children with reading difficulties, children with Down syndrome are older than reading matched control groups and have weaknesses in phonological awareness and decoding. As outlined above, there is some evidence that children with reading difficulties perform more poorly on orthographic learning tasks than would be expected from their reading level, and this may be due to associated phonological difficulties. Therefore it was predicted that children with Down syndrome would show measurable levels of orthographic learning but this would be below their word reading skills. To test this, a larger group of typically developing children were recruited who were expected to be at similar reading accuracy levels to the children with Down syndrome.

If phonological weaknesses are a cause of the poor orthographic learning that we see in children with reading difficulties and are likely to see in children with Down syndrome, then it would be of interest to investigate the effects of phonological pre-training, which has been found to improve orthographic learning in typically developing children (Duff & Hulme, in press; McKague et al. 2001). In relation to typically developing children matched on reading accuracy, it is possible that children with Down syndrome may benefit less or more from phonological pre-training. Phonological awareness is more weakly associated with reading skills in children with Down syndrome than in typical development as shown in Study 1 (see also Boudreau, 2002; Hulme et al., in press). Therefore children with Down syndrome may find it more difficult to apply taught phonological information to the task of decoding a novel word and thus benefit less from phonological pre-training. Conversely, the advantage produced by phonological pre-training may be greater for children with Down syndrome. As this group of children typically find nonword reading difficult, phonological recoding attempts are more likely to be incomplete or unsuccessful. However if children have been provided with an accurate phonological representation of the target word then this may partially match the reading attempt and be selected as the correct pronunciation. Therefore there may be more opportunity for the existing phonological representation to enhance decoding attempts in the group of children with Down syndrome.

It was also hoped that a subgroup of the typically developing children could be pairwise matched to the children with Down syndrome on decoding to test whether orthographic learning was in line with this skill. Children with Down syndrome were predicted to benefit from phonological pre-training to the same extent as this subgroup of typically developing children. If the groups have similar levels of decoding skill then the processes underlying this may be similar, therefore both groups of children will be able to create and apply phonological representations to the task of orthographic learning to the same degree.

To summarise, the current experiment aimed to investigate orthographic learning in children with Down syndrome and test the above predictions. Children with Down syndrome who were able to read took part in this study along with a larger group of typically developing children of a similar reading ability. It was predicted that children with Down syndrome would demonstrate orthographic learning but this would be to a lower extent than the typically developing children. However if a subgroup of the typically developing group could be matched to the children with Down syndrome on decoding, then their levels of performance was predicted to be equivalent. The effect of phonological pre-training was also to be explored.

4.2. Method

4.2.1. Design

Children with Down syndrome and typically developing children took part in an orthographic learning study in which they learnt to read 12 nonwords, half of which received phonological pre-training. Within the same session, phonological choice and orthographic choice post-tests were administered to assess how well the children had learnt the phonology and orthography of the taught words.

4.2.2. Participants

Sixteen children (five boys) with Down syndrome were recruited from the larger sample of children taking part in the longitudinal study. Information about the study was sent to parents of children who were able to read at least one word and had speech which was largely intelligible. All parents or guardians contacted gave informed consent. Children were aged 8 to 17 years and had a mean age of 13;08. According to parental questionnaires, 15 children had Trisomy 21 and one parent did not know what form of Down syndrome their child had. Ten children attended a mainstream school: five in primary and five in secondary. Four children were in special education: two in secondary schools and two in college. Two children had joint attendance at special and mainstream secondary schools.

Thirty typically developing children (16 boys) were recruited from two primary schools in York from Year 1 and Year 2 classes and were aged 5-7 years. The group had a mean age of 6;01. Children who had been identified with special educational needs were excluded. Consent was gained from the headteachers of the schools and also from parents. The children were unselected but the year groups were chosen to correspond to the reading age range of the children with Down syndrome as in Study 2.

4.2.3. Assessment Battery

4.2.3.1. Nonverbal ability.

Matrices was used to assess nonverbal ability as in the previous studies. The typically developing children who took part in this study were all within the standardisation age for the WPPSI-III^{UK} and so were administered the matrices subtest from this test battery. To aid comparison across the two groups, all individuals with Down syndrome were also administered this task. The same administration procedure as Study 1 (section 2.2.3.1.) was followed.

4.2.3.2. Verbal short-term memory.

The WMTB-C word recall subtest was administered to measure verbal short-term memory as in Study 2 (section 3.2.2.3.2.).

4.2.3.3. Reading accuracy.

The single word reading test from the YARC Passage Reading test battery was administered, the details of which are provided in Study 1 (see section 2.2.3.3.3.).

4.2.3.4. Nonword reading.

The GNWRT test was used to test children's decoding skills as in Study 1 (section 2.2.3.3.5.). No extension items were used, hence this followed the same format as at the first time-point in Study 1.

4.2.3.5. Phonological awareness

4.2.3.5.1. Alliteration matching.

Alliteration matching was used to assess phonological awareness as in Study 1 (see section 2.2.3.4.1.).

4.2.3.5.2. Sound deletion.

The sound deletion subtest from the YARC Early Word Reading test battery was also used to assess phonological awareness, as in Study 2 (section 3.2.2.3.4.2).

4.2.3.6. Expressive vocabulary.

The WPPSI-III^{UK} picture naming subtest was administered as in Study 1 (see section 2.2.3.5.1.).

4.2.4. Training Materials

Twelve pairs of nonwords were created which had two different but homophonic vowel digraphs (e.g. 'nirp' and 'nurp'). All nonwords had four graphemes and three phonemes and are shown in Appendix 6. Within the nonword pairs, the nonwords had a similar number and frequency of orthographic neighbours (this was taken from the ARC database; Rastle, Harrington, & Coltheart, 2002). The nonword pairs were separated into two groups ensuring that similar vowel patterns were not all in the same group. Within each pair, nonwords were randomly assigned to be either a target nonword or pseudohomophone distracter, which would be used in the orthographic choice post-test (see section 4.2.6.2.). These assignments were the same for all children. ANOVAs were conducted for the total number and frequency of orthographic neighbours for the four groups of nonwords (i.e. the two groups of target nonwords and two groups of pseudohomophones) and the main effect of group was not significant.

4.2.5. Learning Procedure

All 12 target nonwords underwent orthographic learning and six also received phonological pre-training. The two conditions occurred on different days. The two groups of target nonwords were counterbalanced across the two training procedures (phonological pre-training vs. control), as was the order in which the child experienced the two conditions resulting in four versions of the experiment.

4.2.5.1. Phonological pre-training.

There were four trials in the phonological pre-training, each of which consisted of a repetition and phonological consolidation activity. The task was introduced by telling the children they were going to learn some alien words. First, the children heard the nonword and were asked to repeat it. They then did a phonological consolidation activity, which increased in difficulty throughout the training. For the first trial the children heard the nonword sounded out and had to repeat this and they also heard the first and last sound isolated. The second trial was similar to the first but the children had to sound out the nonword independently. The children had to independently isolate the initial sound and then the final sound for the third and fourth trial respectively. The six nonwords appeared in a fixed random order within each trial and corrective feedback was given.

4.2.5.2. Orthographic learning.

In the control condition the orthographic learning was the first task in the testing session and the nonwords were again introduced as alien words. In the pre-training condition the children were told they were now going to read the alien words. The children saw the written words individually and were asked to read them. They received corrective feedback after each attempt. The words were printed in size 36 Century Gothic lower-case font-type on sheets of A4 paper. All six words appeared in a fixed random order within one trial and there were four trials in total.

4.2.6. Experimental Post-Tests

After an interval of 10-15 minutes, during which the background cognitive tasks were given, children were administered a phonological choice and an orthographic choice post-test. The aim of the phonological choice task was to test whether children had learnt and stored accurate phonological representations for the trained nonwords. The aim of the orthographic choice post-test was to assess children's knowledge of the specific spelling pattern of the nonwords.

4.2.6.1. Phonological choice task.

The phonological choice task was presented on a laptop computer using e-Prime version 1.0. Pictures of three children were shown sequentially, each accompanied by a spoken nonword, which was either the target nonword or a distracter. The three pictures of the children then appeared simultaneously on the computer screen and the child was asked who had said an alien nonword that they had learnt earlier. For each target nonword, two distracters were devised (see Appendix 6). The first differed by one phoneme, which was created by changing one consonant by place of articulation, voicing or manner. For half of the nonwords, the initial letter was changed and for the remaining half, the final letter was changed. The second distracter differed by two phonemes and was created by also changing the remaining consonant in the same way, for example the distracters for 'nirp' were 'nirt' and 'mirt'. All the target nonwords and the distracters were recorded in a soundproof room. Prior to the experimental trials, two practice trials with real target words were administered.

4.2.6.2. Orthographic choice task.

The orthographic choice task was also presented using e-Prime on a laptop computer. Three written nonwords appeared on the computer screen simultaneously and the children were asked to pick the one that they had learnt that day. As outlined above, all target nonwords had a pseudohomophone distracter. A visual distracter was also created by changing the last consonant in the target nonword to a visually similar one, e.g. 'nirg' was the visual distracter for 'nirp'. All distracter items are shown in Appendix 6. Prior to the experimental trials, two practice trials with real words and pictures were administered.

4.2.7. Procedure

4.2.7.1. Typically developing children.

Children took part in two training sessions, which lasted 20-30 minutes each. The orthographic learning procedure took place first in each session, then background tasks were administered, and finally the phonological choice post-test and the orthographic choice post-test. In the first session, matrices, picture naming and single word reading were administered between the training and post-tests. In the second session, the background tasks given were alliteration matching, sound deletion, nonword reading and word recall. Testing took place in a quiet space within the school and children were seen individually. The delay between sessions varied between one and four days, with a mean of 2.17 days.

4.2.7.2. Children with Down syndrome.

This experiment took place at the same time as testing for Time 3 for Study 1, in which all the children with Down syndrome were taking part. All tasks were administered in the same order as the typically developing group. The children with Down syndrome were either assessed in their home or school depending on parental preference. The delay between sessions was typically longer than for the typically developing group due to the logistics of testing in different schools; the mean delay was 9.31 days, and ranged from four to seventeen days.

4.3. Results

Raw scores and an alpha level of .05 were used for all analyses. Data were complete for all measures with the exception of one child with Down syndrome who refused to complete the alliteration matching and sound deletion tasks.

4.3.1. Performance on Background Measures

The two groups were not explicitly matched on any measure but the school year groups of typically developing children were chosen to correspond approximately to the reading ability of the children with Down syndrome. Since single word reading is a relative strength in individuals with Down syndrome it was expected that they would perform at a significantly lower level than the typically developing children on the other measures. The mean scores of the two groups on all measures can be seen in Table 25.

To examine the distribution of scores within the groups the Shapiro-Wilk test was used and histograms, along with measures of skew and kurtosis, were examined. For the typically developing group, a clear ceiling effect was evident for the alliteration matching task and there was a positive skew for word recall. In the data for the group of children with Down syndrome the distribution for picture naming was negatively skewed and the scores for sound deletion showed negative kurtosis. To compare the two groups on these tasks, a Mann-Whitney U task was used. Where the distributions were normal in both groups, then an independent samples t-test was used. All between-group comparisons can be seen in Table 25.

As predicted, there were no differences between the groups on the single word reading task. The two groups also performed similarly on the picture naming task and as such there was no significant difference between them. The typically developing children performed better than the children with Down syndrome on the nonword reading task, and although not significant this difference was marginal. There were significant differences in favour of the typically developing children on the matrices, alliteration matching, sound deletion and word recall tasks.

4.3.2. Phonological Learning

4.3.2.1. Phonological pre-training.

In order to investigate whether the phonological pre-training improved orthographic learning, it must first be established that the pre-training resulted in increased levels of phonological learning in both groups. For the repetition and phonological consolidation aspects of the phonological pre-training, the scores for the two groups are shown in Table 26. The results clearly show that the typically developing group found the pre-training quite easy and were able to correctly say the nonword and demonstrate understanding of its phonology. The children with Down syndrome found both aspects of pre-training significantly harder, although the difference is more striking with the phonological consolidation exercises. This finding is

Table 25.

Scores of the children with Down syndrome and the typically developing children on all background measures (Study 3)

	Scores of children with Down syndrome		Scores of typically developing children		
	Mean (standard deviation)	Range	Mean (standard deviation)	Range	Between-group differences
Matrices (max. 29)	11.81 (4.93)	5-20	15.63 (4.98)	6-24	t(44)=2.49, *p*=.017
Single word reading (max. 60)	26.44 (13.88)	4-45	21.40 (9.08)	8-39	t(22.05)=-1.31, *p*=.204
Nonword reading (max. 20)	6.69 (5.95)	0-17	10.20 (5.49)	1-20	t(44)=2.01, *p*=.051
Alliteration matching (max. 10)	7.20 (2.11)	4-10	9.40 (0.89)	7-10	U=89.00, *p*<.001
Sound deletion (max. 12)	5.00 (3.16)	0-9	8.67 (2.31)	4-12	U=77.00, *p*<.001
Word recall (max. 42)	11.81 (3.12)	7-18	17.77 (4.34)	12-32	U=53.00, *p*<.001
Picture naming (max. 30)	22.13 (4.69)	11-27	23.70 (2.93)	17-29	U=206.50, *p*=.436

Table 26.

Mean scores (and standard deviations) for the children with Down syndrome and the typically developing children on phonological pre-training (Study 3)

	Children with Down syndrome	Typically developing children	Between-group differences
Repetition (max. 24)	21.63 (2.16)	23.70 (0.60)	U=70.50, *p*<.001
Phonological consolidation (max. 24)	13.19 (5.95)	21.50 (2.52)	U=35.00, *p*<.001

unsurprising given the poorer performance of the children with Down syndrome on the background phonological awareness tasks.

4.3.2.2. Phonological choice post-test.

A phonological choice post-test was administered after the orthographic learning procedure to assess how well children could discriminate the taught nonword from phonetically similar distracters. Figure 10 shows the results from the phonological choice post-test. For both conditions and both groups, the correct answer was chosen above chance levels, i.e. two. A mixed ANOVA was conducted for the correct answer with condition (control vs. phonology) as a within-subjects factor and group (Down syndrome vs. typically developing) as a between-subjects factor. There was a main effect of condition, F (1,44)=7.85, *p*=.008, η_p^2=.15, due to better performance in the phonological pre-training condition. Neither the main effect of group, F(1,44)=1.95, *p*=.170, η_p^2=.04, nor the group by condition interaction, F(1,44)=0.94, *p*=.338, η_p^2=.02, were significant. These results illustrate that both groups of children had significantly better phonological representations of the taught words after phonological pre-training compared to orthographic learning only.

4.3.3. Orthographic Learning

4.3.3.1. Orthographic learning procedure.

The scores in the control and phonological pre-training conditions during orthographic learning are shown in Figure 11 for both groups. It can be seen from this that the children with

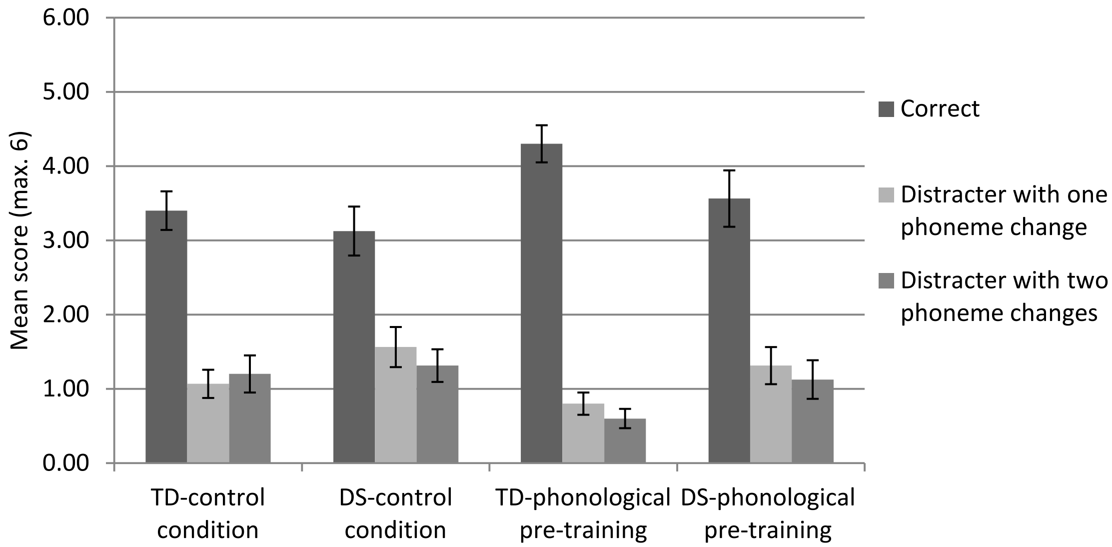


Figure 10. The mean scores for each possible response on the phonological choice post-test for the children with Down syndrome and typically developing children with standard error bars (Study 3)

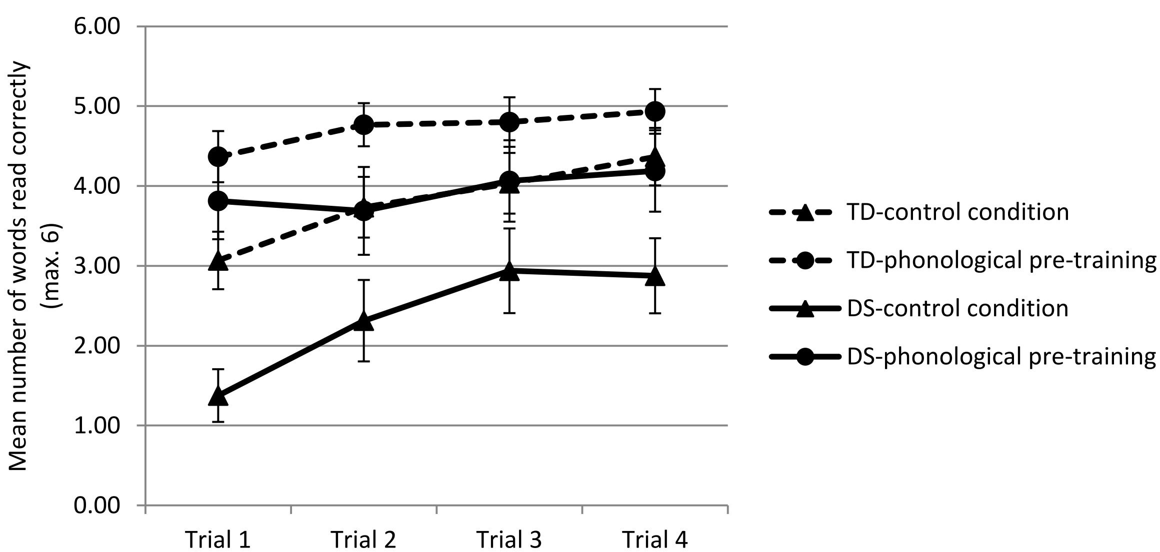


Figure 11. Performance of the typically developing children and children with Down syndrome during orthographic learning with standard error bars (Study 3)

Down syndrome generally performed at a lower level than the typically developing children. A clear advantage for both groups across the learning procedure in the phonological pre-training condition compared to the control condition can also be seen. A maximum of 24 words could be read correctly during training. The typically developing group read a mean of 15.20 words correctly in the control condition and 18.87 in the phonology condition overall. The group of children with Down syndrome read a mean of 9.50 words correctly in the control condition and 15.75 in the phonology condition. Figure 11 also indicates that learning increased over the training procedure, although this appeared to be greater in the control condition.

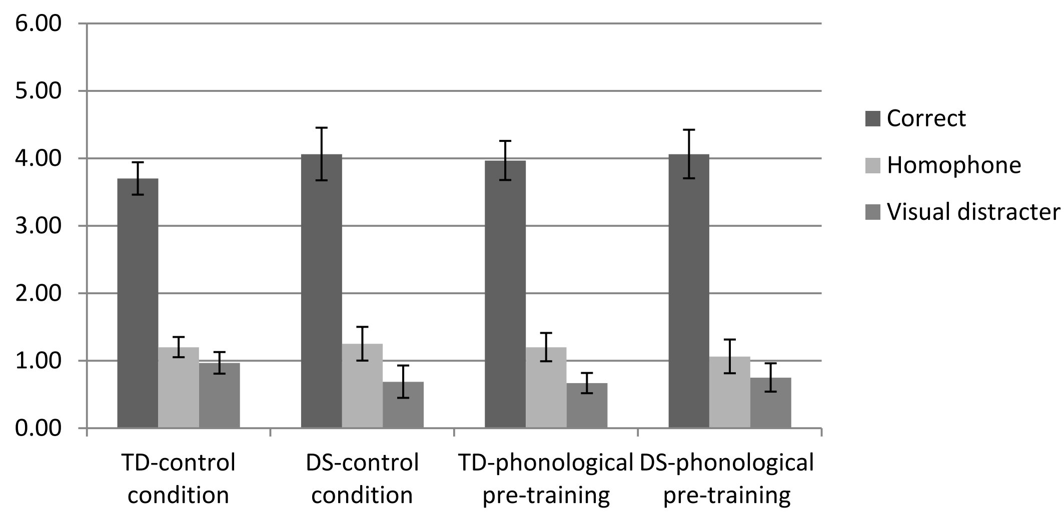
A mixed ANOVA with condition (control vs. phonological pre-training) and trial (1-4) as within-subject variables and group (Down syndrome vs. typically developing) as a between-subject variable was conducted. There was a main effect of condition, with the phonological pre-training condition yielding significantly higher scores than the control condition, $F(1,44)=59.44$, $p<.001$, $\eta_p^2=.58$. There was also a main effect of group as the typically developing children scored significantly higher than the children with Down syndrome, $F(1,44)=4.50$, $p=.040$, $\eta_p^2=.09$, and a main effect of trial as scores improved during the training, $F(3,132)=25.71$, $p<.001$, $\eta_p^2=.57$. Further, there was a significant interaction between condition and trial, $F(3,132)=6.89$, $p<.001$, $\eta_p^2=.26$, which was due to greater improvement across trials in the control condition. The other two-way and three-way interactions were not significant although the condition by group interaction was marginal, $F(1,44)=4.03$, $p=.051$, $\eta_p^2=.08$. Figure 11 illustrates that this is due to a greater discrepancy between the scores in the two conditions for the children with Down syndrome.

The marginally significant interaction was followed up with a Tukey's HSD test. A HSD value of 2.44 was obtained, which showed that both groups performed better in the pre-training condition compared to the control condition and the typically developing group performed better than the children with Down syndrome in both conditions. However the real comparison of interest is comparing the difference between the two conditions across groups. The difference between the two conditions in the typically developing group was 3.67, $d=0.54$, and 6.25, $d=0.84$ for the group with Down syndrome. The effect size is medium in the typically developing group and large for the children with Down syndrome, indicating a greater difference between the two conditions for the children with Down syndrome.

4.3.3.2. Orthographic choice post-test.

An orthographic choice post-test was carried out after the orthographic learning procedure to investigate whether children could discriminate the target nonword from a pseudohomophone and a visual distracter. Figure 12 shows the performance of both groups on the orthographic choice task. It can be seen that the correct answer was consistently chosen at above chance levels, i.e. two. Both the pseudohomophone and visual distracter were chosen at similarly low levels by both groups and in both conditions. A mixed ANOVA on correct answers with condition (control vs. phonology) as a within-subject factor and group (Down syndrome vs. typically developing) as a between-subjects factor was carried out and as Figure 12 suggests no main effect or interaction were significant.

Figure 12. The mean scores for each possible response on the orthographic choice post-test for the children with Down syndrome and typically developing children with standard error bars (Study 3)



4.3.3.3. Summary of results.

The phonological pre-training resulted in more successful performance on the phonological choice post-test and most importantly led to greater accuracy during orthographic learning than no pre-training. The children with Down syndrome showed poorer performance during orthographic learning than the typically developing children, although there was a marginally significant interaction for the children with Down syndrome to benefit more from the phonological pre-training. During the orthographic learning, there was an overall improvement in accuracy and this was greater for the words which had not received phonological pre-training. However there was no effect of phonological pre-training or group when identifying the correct spelling pattern of the target nonword.

4.3.4. Matching for Target Nonword Decoding Skill

The unselected group of typically developing children were well matched to the children with Down syndrome on reading accuracy, although the typically developing group performed significantly better during orthographic training. The trend for a greater advantage for the phonological pre-training condition in the children with Down syndrome may be due to their being less able to decode the nonwords in the control condition and therefore having more room for improvement in the pre-training condition. Therefore a subgroup of typically developing children were pairwise matched to the children with Down syndrome on their scores on the first orthographic training trial in the control condition. The children were either matched on their exact score or within one point. This resulted in 16 typically developing children (seven males) being selected who had a mean age of 6;02 and the full sample of 16 children with Down syndrome remained.

4.3.4.1. Performance on background measures.

The performance of the typically developing subgroup on the background cognitive measures is shown in Table 27 along with the children with Down syndrome for comparison purposes. Matching on target decoding also results in the groups having similar scores on the background nonword reading test. Due to the uneven profile in individuals with Down syndrome with reading accuracy generally being a strength compared to nonword reading, the children with Down syndrome performed significantly better on the reading accuracy measure than the typically developing subgroup. The two groups had similar scores on the picture naming task, but the typically developing subgroup performed significantly better on the remaining tasks of phonological awareness, nonverbal ability and verbal short-term memory.

Table 27.

Scores on all background measures for the children with Down syndrome and the typically developing subgroup pairwise matched on target decoding (Study 3)

	Scores of children with Down syndrome		Scores of typically developing children		
	Mean (standard deviation)	Range	Mean (standard deviation)	Range	Between-group differences
Matrices (max. 29)	11.81 (4.93)	5-20	15.69 (4.84)	6-23	t(30)=2.24, *p*=.032
Picture naming (max. 30)	22.13 (4.69)	11-27	23.75 (2.89)	18-29	U=110.50, *p*=.507
SWRT (max. 60)	26.44 (13.88)	4-45	16.31 (7.74)	8-37	t(23.52)=-2.55, *p*=.018
Alliteration matching (max. 10)	7.20 (2.11)	4-10	9.25 (1.06)	7-10	U=51.50, *p*=.005
Sound deletion (max. 12)	5.00 (3.16)	0-9	7.56 (2.31)	4-11	U=65.50, *p*=.030
Nonword reading (max. 20)	6.69 (5.95)	0-17	6.50 (3.03)	1-14	U=123.00, *p*=.850
Word recall (max. 42)	11.81 (3.12)	7-18	18.25 (4.73)	12-32	t(30)=4.55, *p*<.001

4.3.4.2. Phonological learning.

4.3.4.2.1. Phonological pre-training.

As with the previous analysis, scores were examined on the phonological pre-training exercises and these are shown in Table 28. The subgroup of typically developing children performed similarly to the overall typically developing sample and therefore significant differences between them and the children with Down syndrome on both the repetition and phonological consolidation aspects of pre-training were present.

Table 28.

Mean scores (and standard deviations) for the group of children with Down syndrome and the typically developing sub group on phonological pre-training (Study 3)

	Group with Down syndrome	Typically developing subgroup	Between-group differences
Repetition (max. 24)	21.63 (2.16)	23.81 (0.40)	U=31.50, *p*<.001
Phonological consolidation (max. 24)	13.19 (5.95)	20.88 (2.94)	U=27.00, *p*<.001

4.3.4.2.2. Phonological choice post-test.

Figure 13 shows the scores for the phonological choice post-test, which are similar to the results with the whole typically developing control group. The mean score for the correct answer is above chance, i.e. two, across both conditions and groups, but this is higher in the pre-training condition in both groups. Indeed a mixed ANOVA found a main effect of condition, $F(1,3)=5.77$, $p=.023$, $\eta_p^2=.16$, due to higher scores in the pre-training condition, but the main effect of group and group by condition interaction were not significant. The results from this post-test confirm that the phonological pre-training resulted in better phonological representations of the target nonwords in both groups.

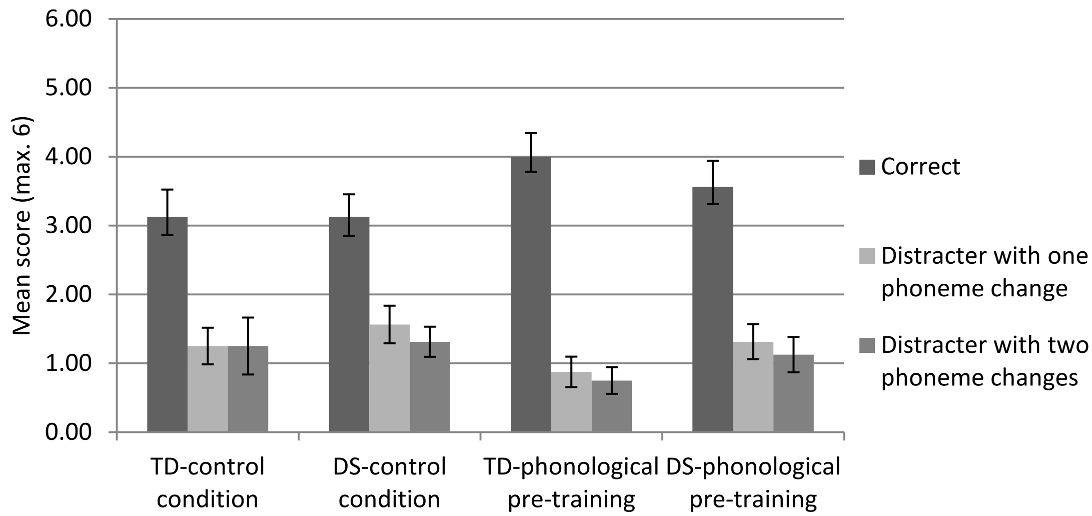


Figure 13. The mean scores for each possible response on the phonological choice post-test for the children with Down syndrome and typically developing subgroup with standard error bars (Study 3)

4.3.4.3. Orthographic learning.

4.3.4.3.1. Orthographic learning procedure.

The scores in the control and phonological pre-training conditions during orthographic learning are shown in Figure 14 for both groups. It can be seen from this that the levels of learning were similar in the two groups in both conditions. Furthermore there was a clear benefit for nonwords which received phonological pre-training, and this is to the same extent in the two groups. A maximum of 24 words could be read correctly during training. The typically developing subgroup read a mean of 10.12 words correctly in the control condition and 15.63 in the phonology condition overall. As reported above, the group of children with Down syndrome read a mean of 9.50 words correctly in the control condition and 15.75 in the phonology condition. As with the analysis with the whole sample of typically developing children, learning increased over the training procedure, but this appeared to be greater in the control condition.

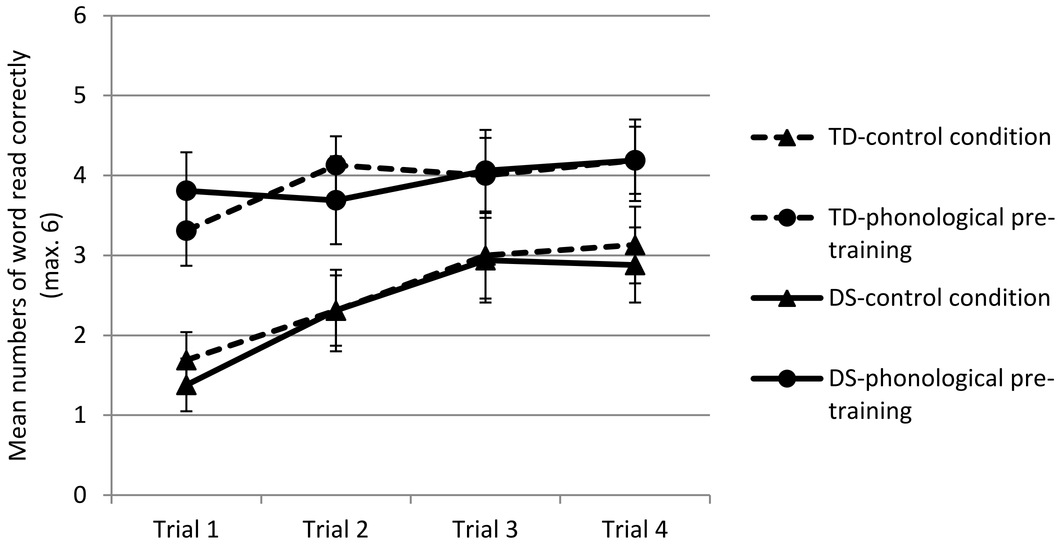


Figure 14. Performance of the children with Down syndrome and typically developing subgroup during orthographic training with standard error bars (Study 3)

In a mixed ANOVA, there was a main effect of condition, due to higher scores in the phonological pre-training condition, $F(1,30)=56.84$, $p<.001$, $\eta_p^2=.66$. Due to the improvement over training, the main effect of trial was significant, $F(3,90)=22.06$, $p<.001$, $\eta_p^2=.63$. The main effect of group was not significant, $F(1,43)=0.01$, $p=.915$, $\eta_p^2=.01$. The only interaction which was significant was condition by trial, $F(3,90)=4.38$, $p=.006$, $\eta_p^2=.24$, due to the greater improvement in the control condition across trials.

4.3.4.3.2. Orthographic choice post-test.

The results for the orthographic choice post-test are shown in Figure 15. The mean number of correct answers was above chance, i.e. two, for both groups in both conditions. The children with Down syndrome appeared to be performing at a slightly higher level than the typically developing subgroup. However there was no significant main effect of group or condition or an interaction between the two in a mixed design ANOVA.

4.3.4.4. Summary of results.

When the two groups were matched for target decoding they performed equivalently during the orthographic learning. Both groups benefited from phonological pre-training, and

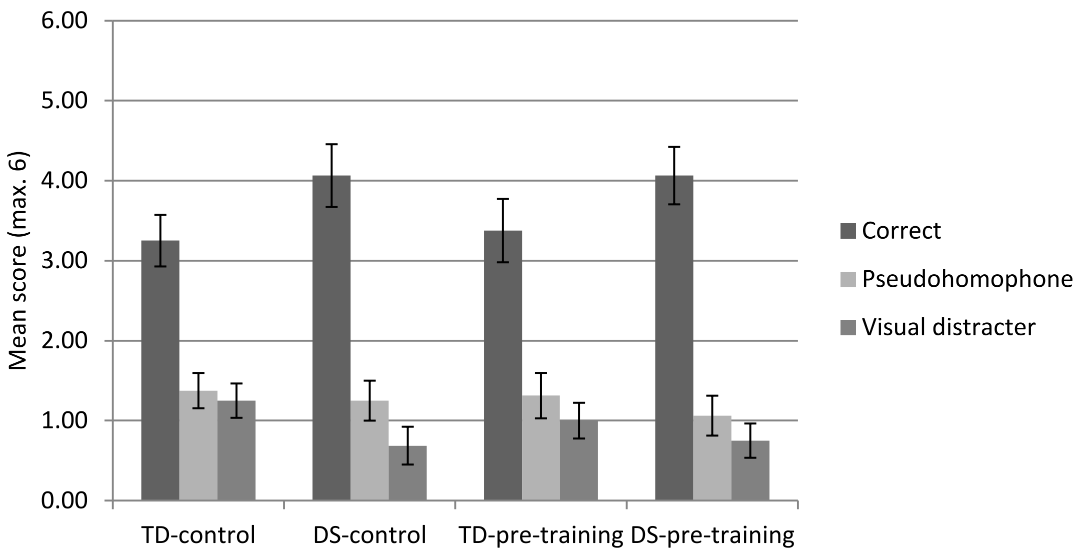


Figure 15. The mean scores for each possible response on the orthographic choice post-test for the children with Down syndrome and typically developing subgroup with standard error bars (Study 3)

this is the same across the learning procedure. As with the analyses with the whole sample of typically developing children, there was no benefit of phonological pre-training on the orthographic choice post-test.

4.3.5. Relationship between Cognitive Skills and Orthographic Learning

To examine which cognitive measures were related to orthographic learning, correlations were computed. The total learning scores from the control and phonological pre-training conditions were correlated strongly in both groups (r=.77** for the children with Down syndrome and r=.88** for the typically developing children) and so orthographic learning across both conditions was summed to yield a total orthographic learning score.

The simple correlations are shown in Table 29. It can be seen that picture naming, single word reading, phonological awareness, nonword reading and word recall are all correlated with orthographic learning at a moderate to high level in the group of children with

Table 29.

Simple and partial correlations controlling for reading ability between orthographic training measure and background measures for the children with Down syndrome and typically developing children(Study 3)

	Simple correlations		Partial correlations (controlling for reading)	
	Children with Down syndrome	Typically developing children	Children with Down syndrome	Typically developing children
Age	-.11	.19	.03	-.26
Matrices	.25	.35[a]	.05	.26
Picture naming	.84**†	.39* †	.32	.12
Word reading	.84**†	.76** †	-	-
Alliteration matching	.51[b]	.50**	.39	.41*
Sound deletion	.62*	.59**	-.05	.23
Nonword reading	.78**	.68**	.19	.15
Word recall	.77** †	.17 †	.15	-.20

Note. [a]p=.054 [b]p=.059
† denotes a significant between-group difference in correlation strength
*p<0.05 **p<0.01

Down syndrome. Of note is the particularly high correlation with word reading. In the typically developing group, the correlations with cognitive measures are generally significant but at a lower level than in the group with Down syndrome. Word recall however is not significantly related to orthographic learning. A number of the cognitive tasks had significantly stronger correlations with orthographic learning in the children with Down syndrome, namely picture naming, single word reading and word recall.

In contrast to Studies 1 and 2, nonverbal ability and age were not controlled for in a partial correlation as the relationships between these variables and orthographic learning were generally weak in the two groups. Due to the high correlation with reading skill in both groups, this was entered as a control variable in a partial correlation. The aim of this was to investigate if any other variables were related to orthographic learning beyond the effects of reading. These partial correlations are shown in Table 29. The correlations in the group of children with Down syndrome reduced to a non-significant level, which is likely due to the very strong correlation between orthographic learning and reading accuracy. However, picture naming and alliteration matching were still moderately correlated. In the typically developing group, alliteration matching was the only variable which remained significantly correlated with orthographic learning. The strength of the correlation was very similar to that for the children with Down syndrome but presumably was not significant in this group due to the smaller sample size.

4.4. Discussion

Study 3 examined orthographic learning in children with Down syndrome, and specifically the effect of phonological pre-training. The first aim of this experiment was to investigate whether children with Down syndrome showed a deficit during orthographic learning relative to a control group of typically developing children. The two groups of children were matched on reading accuracy but not nonword reading and thus the typically developing children performed significantly better during the orthographic learning procedure. When a subgroup of the typically developing children was matched to the group of children with Down syndrome on target decoding skill, this group difference no longer remained. The second aim of this experiment was to explore whether phonological pre-training improved orthographic learning, and whether this was to a similar extent in both groups. It was found that phonological pre-training did benefit performance during orthographic learning, and this was similar in the children with Down syndrome and typically developing children when matched on target decoding. However, this effect was not seen on an orthographic choice post-test.

The typically developing children read more nonwords correctly during orthographic learning than the children with Down syndrome. As the typically developing children had better existing nonword reading skills, the two groups were not equated for baseline performance in the experiment. Therefore 16 of the typically developing children were pairwise matched to the children with Down syndrome on their scores from the first trial of learning in the control condition. This also resulted in the two groups being well matched on the standardised measure of nonword reading. There were no significant differences between the typically developing subgroup and the children with Down syndrome during orthographic learning. Thus as predicted, on the task of orthographic learning the children with Down syndrome performed in line with their nonword reading ability but more poorly than would be expected from their reading accuracy. Similarly, studies with children with dyslexia have found that they also show levels of orthographic learning below their reading accuracy ability (Bailey et al., 2004; Ehri & Saltmarsh, 1995; Reitsma, 1983).

The phonological pre-training exposed children to the spoken nonwords and required them to engage in activities that emphasised the individual phonemes within the nonwords. Pre-trained nonwords were subsequently better identified from similar sounding nonwords on a phonological choice post-test by both groups, suggesting that the pre-training was successful. As predicted, nonwords that received the phonological pre-training were read more accurately. Having a pre-existing phonological representation of a word may aid children by providing a match to a partial decoding attempt. The nonwords in this study were regular but included vowel digraphs, which can be a source of error for young readers (Share, 1995). During this experiment some children from both groups did attempt to pronounce the vowel digraphs as individual graphemes, often resulting in partial decoding attempts. It is proposed that these partial decoding attempts will activate the correct phonological representation, which is then selected as the pronunciation.

Both groups of children benefited from phonological pre-training during orthographic learning. There was a marginally significant effect for the children with Down syndrome to benefit more from the pre-training than the typically developing children at the same level of reading. It may be because the group of children with Down syndrome had poorer performance in the control condition than the typically developing children, therefore there was simply more potential for a difference between the two conditions. Furthermore as the children with Down syndrome were less accurate overall, they may have produced more partial decoding attempts. For pre-trained words, this may then activate the newly-created phonological representation. In contrast, as the typically developing children were more accurate, there would be less opportunity for a benefit of phonological pre-training. The between-group difference was no longer present when the two groups of children were matched for target decoding. This may be because the groups now make an equal number of unsuccessful decoding attempts across the groups, leading to an equivalent potential for benefit from the phonological pre-training.

The accuracy of both typically developing children and children with Down syndrome increased throughout the orthographic learning procedure. However the improvement was greater in the control condition than the phonological pre-training condition. A possible reason may be that the children were more accurate on the first trial for the pre-trained nonwords than for the control nonwords, therefore there was less potential for growth across trials. The nature of the corrective feedback during orthographic learning may also have contributed to this difference between conditions. For the feedback, children heard the correct pronunciation of the target nonword. At the beginning of the orthographic learning procedure, children had not yet heard the phonology of the control nonword, whereas they had for the pre-trained nonwords. Therefore the feedback may have provided more benefit to the control nonwords as it added new phonological information to the lexical representation. In contrast the feedback did not provide any new information for the pre-trained nonwords and therefore would not be expected to provide as much benefit.

Established orthographic representations are often assessed using an orthographic choice task, which tests whether children are able to identify the taught orthographic form when also presented with a pseudohomophone. Importantly this task cannot be completed using decoding strategies. Previous studies which have compared the effect of phonological pre-training have not included post-tests that specifically test orthographic representations, i.e. orthographic choice and naming in comparison to pseudohomophones or spelling (Duff & Hulme, in press; McKague et al., 2001). In the present experiment the target item was picked at relatively high levels of accuracy in an orthographic choice post-test by both groups of children and in both conditions. Therefore the same level of orthographic knowledge was demonstrated for nonwords that received phonological pre-training and those that did not. It could be that repeated exposure to the orthography of the target word resulted in children creating and storing an orthographic representation of the nonword, regardless of whether they read it correctly. To test the unique contribution of phonology, rather than the combined effect of phonological and orthographic exposure, the orthographic choice could be administered after the first trial of orthographic learning. Furthermore the task may not be sensitive enough to effects of group or pre-training as children performed relatively accurately. If more items were trained then the potential for differences between the two conditions, and groups, would increase.

The correlations of the background cognitive measures with performance during the orthographic learning procedure were generally higher for the group of children with Down syndrome, particularly for verbal short-term memory and vocabulary. The test of existing reading skill assessed acquired orthographic representations and as would be expected this was highly correlated with orthographic learning in both groups, although again this was stronger in the children with Down syndrome. The relationship of orthographic learning with phonological awareness and nonword reading was moderate in both groups, which may suggest that both groups utilise these skills to a similar extent in orthographic learning. In support of this, when reading was controlled for the relationship between alliteration matching and orthographic learning remained moderate in both groups. This highlights the importance of phonological awareness in orthographic learning, as the self-teaching hypothesis suggests. Share (1995) highlighted factors such as print exposure and visual memory which may also affect orthographic learning. As these two factors are often relatively advanced in Down syndrome, due to age and their cognitive profile, it would be interesting to examine these correlates in children with Down syndrome and compare this to typically developing children.

It is argued that phonological pre-training improved orthographic learning because a phonological representation of the nonword was created and this was accurate and strong enough to be matched to a partial decoding attempt and then be produced. However this study cannot address the nature of phonological information that is needed for this to occur. McKague et al. (2001) found that hearing and repeating a word compared to only hearing it resulted in similar benefits on orthographic learning for typically developing children. This suggests that detailed information about a word’s phonology, i.e. its individual phonemes, or even the production of it, is not necessary to produce benefits. Further research is needed to investigate how in-depth the phonological information needs to be for children with Down syndrome.

A future avenue for research could be to examine the effect of semantic pre-training on orthographic learning. This has been examined in typically developing children and there tends to be little advantage for semantic and phonology pre-training over phonology only pre-training (Duff & Hulme, in press; McKague et al., 2001). There is some evidence however, that semantics may be more important when the phonology to orthography mappings are weak, for example for words with inconsistent pronunciations (McKay, Davis, Savage, & Castles, 2008; Taylor, Plunkett, & Nation, 2011). In context of the triangle model of reading, this would result in reduced efficiency of the phonology-orthography pathway and therefore the influence from the indirect pathway via semantics may be more important. This may also be true for children who have phonological deficits such as children with Down syndrome.

4.4.1. Summary and Conclusions

This experiment has demonstrated that children with Down syndrome show levels of orthographic learning commensurate to their decoding ability. They also benefited from phonological pre-training to the same extent as typically developing children. It is likely then that during reading instruction children with Down syndrome, along with typically developing children, would benefit from being familiar with the phonological form of the word prior to seeing its orthography. Future research can inform teaching and practice further by clarifying what aspect of the phonological pre-training is necessary to produce this advantage.

Chapter Five

General Discussion

5.1. Research Background and Aims

Individuals with Down syndrome have an uneven cognitive profile with relative strengths in reading accuracy and visual memory but comparative weaknesses in phonological awareness, decoding, reading comprehension, verbal memory and oral language (Buckley, 1995; Jarrold et al., 1999; Nash & Heath, 2011; Roch & Jarrold, 2008; Rondal, 1995). Despite being a strength in children with Down syndrome, the development of reading accuracy does not appear to be influenced by the same oral language skills as typically developing children. Specifically, phonological awareness has been found to play a weaker role in reading accuracy, while there is some suggestion that broader oral language skills play a greater role (e.g. Hulme et al., in press).

Learning to read has been suggested to promote oral language development in children with Down syndrome (Buckley, 1995). Case studies with young children with Down syndrome have suggested that introducing words in their written form promotes their emergence in speech (de Graaf, 1993; Duffen, 1976). However this has not been tested experimentally or in comparisons with typically developing children. There are also longitudinal studies which address the effect of learning to read on different aspects of oral language but their findings are mixed partly due to methodological differences such as whether the autoregressor is controlled for (Cupples & Iacono, 2002; Hulme et al., in press; Laws et al., 1995; Laws & Gunn, 2002).

As reading accuracy levels reached by children with Down syndrome increase, their reading comprehension is becoming the focus of more studies. Compared to reading accuracy, reading comprehension is a relative weakness for individuals with Down syndrome (e.g. Nash & Heath, 2011; Roch & Levorato, 2009). The simple view of reading proposes that reading comprehension is the product of decoding and linguistic comprehension (Gough & Tunmer, 1986) and it has been suggested that for individuals with Down syndrome, oral language may have a stronger relationship with reading comprehension than in typically developing children

(Roch & Levorato, 2009). However this may be due to different levels of reading accuracy in the groups of children with Down syndrome and typically developing children (Nash & Heath, 2011). Furthermore there is a lack of longitudinal studies which explore predictors of reading comprehension in children with Down syndrome.

The broad aim of this thesis was to explore the interaction between reading and oral language skills in children with Down syndrome. Study 1 examined the relative longitudinal contributions of phonological awareness and broader oral language skills to the development of reading accuracy. This study also investigated whether reading accuracy promoted oral language development. Finally Study 1 also aimed to evaluate reading comprehension development in line with the simple view of reading. Studies 2 and 3 utilised word learning paradigms to examine the reciprocal relationship between reading accuracy and oral language skills. In Study 2c, children were taught spoken nonwords with or without their orthographic form present to test whether this benefited the production of its spoken form in children with Down syndrome to a greater extent than typically developing children. Study 3 examined the effect of phonology on learning to read new words. Children were taught to read nonwords, half of which received phonological pre-training and the benefit of this for children with Down syndrome relative to typically developing children was examined.

5.2. The Effect of Phonological and Broader Oral Language Skills on Reading Accuracy

5.2.1. Predictors of Reading Accuracy Development

Study 1 examined the longitudinal relationships between reading accuracy, reading comprehension and oral language skills. A group of 21 individuals with Down syndrome were assessed at three time-points, along with a group of 29 typically developing children who were matched for reading accuracy at the outset of the study. Here the results regarding the effect of phonological and non-phonological oral language skills on reading accuracy will be considered.

Path models examined the relative contributions of Time 1 reading accuracy, phoneme awareness and vocabulary to Time 3 reading accuracy. Initial levels of reading accuracy were a strong predictor of Time 3 reading accuracy in both groups of children, but significantly more so in the children with Down syndrome, which is likely to be due to the lower levels of progress in this group. This replicates the findings of Hulme et al. (in press) and highlights the longitudinal stability of reading accuracy in both typically developing children and children with Down syndrome. As the autoregressive effect was so strong, phoneme awareness and vocabulary did not predict variance in later reading in either group. In future longitudinal studies spanning a wider time frame should be conducted with individuals with Down syndrome, as this should result in more progress in reading accuracy.

In path models with phoneme awareness and vocabulary as concurrent predictors of reading accuracy, vocabulary emerged as a unique predictor for the children with Down syndrome whereas neither variable significantly predicted reading accuracy for the typically developing children, although there was a trend for phoneme awareness to be significant. From previous research (e.g. Bradley & Bryant, 1983; Muter, 1994; Muter et al., 2004; Stuart & Masterson, 1992; Wagner et al., 1997), the relationship between phoneme awareness and reading accuracy was expected to be significant in the typically developing group and therefore it is likely there are methodological reasons for this unexpected result. Possible reasons for this include the sensitivity of the task for the typically developing children, the effect of controlling for age and the nature of the phoneme awareness task used. These are discussed in Chapter 2 in more detail but the latter explanation will be highlighted here as it may also have implications for the results for the children with Down syndrome. The phoneme awareness measure used in this study was an alliteration matching task in which children were presented with a target item and two possible responses and were required to select which one started with the same sound as the target item. If a child was familiar with the words their pre-existing lexical representations may have supported their performance on this task. Therefore this may have resulted in shared variance with vocabulary knowledge. In future, different phoneme awareness tasks with lower vocabulary demands should be used to investigate whether phoneme awareness has separable effects from vocabulary on reading accuracy.

The concurrent findings of a weak relationship between phoneme awareness and reading accuracy and a significant relationship between vocabulary and reading accuracy in children with Down syndrome is in line with Hulme et al. (in press). It should be noted here that there was some degree of association between phoneme awareness and reading accuracy and other studies have found this relationship to be significant in children with Down syndrome (Fowler et al., 1995; Gombert, 2002; Kennedy & Flynn, 2003; Snowling et al., 2002). The weight of evidence seems to suggest that although there may be a relationship between phoneme awareness and reading accuracy in children with Down syndrome, this is weaker compared to what would be expected in typically developing children.

Hulme et al. (in press) proposed that the effect of phoneme awareness on reading accuracy may be more variable in children with Down syndrome than in typically developing children. They suggested that some children may develop sufficient phonemic skills to form, and presumably utilise, connections between orthography and phonology, whereas others may depend more on less segmented lexical representations for reading, which may be at the whole-word level. To test this, a larger sample of individuals with Down syndrome would be needed to investigate the existence of a sub-group of children who have good phoneme awareness skills that are related to reading accuracy to a similar extent as in typical development.

Vocabulary was a significant concurrent predictor of reading accuracy in the children with Down syndrome but not in the typically developing children. As the vocabulary tasks in this study tapped phonological representations more than semantics, it is suggested that knowledge of the phonological form of a word helps children with Down syndrome to resolve partial decoding attempts for unknown words. This point will be returned to below in section 5.2.3.

Considering the vocabulary tasks separately, both were moderately and significantly correlated with later reading accuracy for the typically developing children. However for the children with Down syndrome, the correlation with receptive vocabulary was not significant and the correlation with picture naming was strong and significant. Although there were no significant differences between the two groups, this pattern of results is potentially interesting and merits consideration. If the relationship between vocabulary and reading accuracy in the children with Down syndrome is due to the activation of pre-existing phonological representations, then picture naming is a stricter test of the quality of these representations and this may be why it is more strongly correlated with reading accuracy than receptive vocabulary.

An aim of this study was to extend the findings of Hulme et al. (in press) and examine the relationship of reading accuracy with a wider range of oral language tasks. Simultaneous regression path models could not be used for this due to relatively small sample sizes. Therefore longitudinal correlations controlling for age and nonverbal ability were examined, the results of which were similar to the Time 1 concurrent correlations. In both groups, nonword repetition was correlated with later reading accuracy but not when the autoregressor was controlled for. Indeed, a recent study has suggested that nonword repetition may be a consequence rather than cause of reading development (Nation & Hulme, 2011). Considering the grammar measures, receptive syntax was significantly correlated with reading accuracy in the typically developing group only. As children with Down syndrome had lower levels of syntactic knowledge, this may be less likely to aid word reading by providing syntactic constraints on an unfamiliar word. MLU was a significantly stronger correlate of later reading accuracy in the group of children with Down syndrome compared to the typically developing children, although this may not be independent of expressive vocabulary (Dethorne et al., 2005; Devescovi et al., 2005). The narrative task administered to gain a measure of MLU also provided a narrative content score which reflected how well children could describe the key events of the story, but this did not appear to predict later reading accuracy in either group.

5.2.2. The Effect of Phonological Pre-Training on Learning to Read New Words

Study 1 examined the relationship between measures of existing reading and oral language skills whereas Study 3 investigated the effect of phonological pre-training when learning to read new words. Sixteen children with Down syndrome and 30 typically developing children were taught to read nonwords, half of which received phonological pre-training. There were two outcome measures: orthographic learning trials and an orthographic choice post-test. During the orthographic learning procedure, children were effectively being asked to decode the nonwords. In contrast, the orthographic choice post-test measured newly acquired orthographic representations and could not be completed using decoding strategies.

The typically developing group were chosen to be at a similar reading level to the group of children with Down syndrome to determine whether there was a discrepancy between their acquired reading skill and the rate at which new orthographic representations are acquired. Like children with dyslexia (e.g. Bailey et al., 2004; Ehri & Saltmarsh, 1995; Reitsma, 1983), the children with Down syndrome performed significantly worse than typically developing children during orthographic learning. Share (e.g. Jorm & Share, 1983; Share, 1995) proposed that orthographic representations are created by self-teaching, which is mostly dependent on phonological recoding, a process by which graphemes (or larger orthographic units) are recoded into phonemes (or larger phonological units). As the children with Down syndrome performed more poorly on background tasks of nonword reading and phonological awareness, they are likely to be less accurate during phonological recoding. The children with Down syndrome were older than the typically developing children and it may be that they require more exposures to acquire new orthographic representations than typically developing children. This could be investigated by training children to criterion and examining whether children with Down syndrome require more trials.

During the orthographic learning procedure children were being trained to read nonwords. This is therefore akin to a task of decoding, a skill in which children with Down have relative difficulties. A subgroup of typically developing children was matched to the children with Down syndrome on scores from the first orthographic learning trial in the control condition, and there were no differences between the two groups during orthographic learning in either condition. The two groups were also matched on a background measure of nonword reading, and therefore these results suggest that children with Down syndrome are able to acquire, store and retrieve new orthographic representations at a level expected from their decoding ability.

The nonwords used in this study had regular grapheme-phoneme correspondences and it would be of interest to investigate whether individuals with Down syndrome also have relative difficulties with learning to read irregular words. These words would be expected to rely less on phonological recoding and therefore children with Down syndrome may show less difficulty during orthographic learning relative to their reading accuracy level. Indeed Bailey et al. (2004) found that children with dyslexia who performed significantly worse than a reading accuracy matched control group on background tasks of nonword reading and phonological awareness also had relative difficulties learning to read new regular but not exception words.

The phonological pre-training is argued to benefit orthographic learning by providing children with a phonological representation of the new word and therefore a pronunciation match for partial decoding attempts, which results in more accurate responses. This is particularly relevant when children cannot decode the new word readily. Although the nonwords in this study were given regular pronunciations, they included long vowels and children were in the early stages of literacy development. Therefore perhaps the task of decoding was relatively effortful and thus resulted in an opportunity for phonological familiarity to produce a benefit.

These results are also in keeping with the lexical quality hypothesis (Perfetti & Hart, 2002), which states that a high-quality lexical representation incorporates phonological, semantic and orthographic information, all of which are activated when the word is encountered. Furthermore the multiple representations are thought to result in a strong and consistent core representation. Nonwords that underwent phonological pre-training in the present study are more likely to have a stronger lexical representation, resulting in increased accuracy when reading them.

Familiarity with the phonological form of words prior to learning to read them has been found to increase accuracy in typically developing children (Duff & Hulme, in press; McKague et al., 2001; Share, 2004), but this has not yet been examined in children with phonological difficulties. When children with Down syndrome and typically developing children were matched for reading accuracy (but not decoding), there was a trend for children with Down syndrome to benefit more from phonological pre-training during orthographic learning. This is likely to be because children with Down syndrome made more partial decoding attempts, thus there was more opportunity for pre-existing phonological representations to benefit reading. Compared to the subgroup of typically developing children matched on target decoding, the children with Down syndrome benefited to the same extent from phonological pre-training. This is because the two groups have equivalent baseline performance and therefore there is the same opportunity for phonological pre-training to produce an advantage.

Orthographic learning tested how accurately children could read the nonwords. The orthographic post-test assessed children's ability to discriminate the taught spelling pattern from a pseudohomophone, therefore to complete this successfully children needed to have an established orthographic representation. There was no difference on this measure between the children with Down syndrome and the typically developing children when matched on reading accuracy or target decoding. Furthermore, in contrast to the orthographic learning procedure, there was no effect of pre-training. As discussed in Chapter 4, this may be due to equivalent exposure to the orthographic form of the word in the two training conditions or a lack of sensitivity in this task.

5.2.3. The Effect of Lexical Phonology on Reading Accuracy Development

Studies 1 and 3 investigated the effect of oral language skills on reading accuracy in children with Down syndrome. Study 1 employed a longitudinal design to do this, in which the relationships between acquired levels of reading accuracy, phonological awareness and broader oral language skills were considered, both concurrently and longitudinally. Neither phoneme awareness nor vocabulary predicted reading accuracy longitudinally in children with Down syndrome, but vocabulary was a significant concurrent predictor of reading accuracy whereas phoneme awareness was not. Study 3 utilised a dynamic assessment of the relationship between phonology and reading accuracy with an orthographic learning task. Children with Down syndrome benefited to the same extent as typically developing children matched on target decoding from phonological pre-training. This may at first seem in contrast with the results of Study 1 but let us consider the nature of the vocabulary tasks in Study 1 and the phonological pre-training in Study 3.

The vocabulary tasks employed in Study 1 might be argued to be more dependent on phonological than semantic representations as they could be completed with very little semantic knowledge. Indeed Nation and Cocksey (2009) suggest that it is the phonological element of vocabulary knowledge, i.e. lexical phonology, which accounts for the contribution of vocabulary tasks to reading accuracy by helping to resolve partial decoding attempts. It is suggested that in Study 3, as they had poorer nonword reading skills, the children with Down syndrome may have made more partial decoding attempts than the typically developing children. These incomplete attempts may then be resolved by the activation of a matching phonological representation.

In Study 1, the contribution of phonological and semantic aspects of vocabulary knowledge to reading accuracy cannot be separated. To further investigate which aspects of vocabulary knowledge are related to reading accuracy in children with Down syndrome, lexical decision and definitions tasks using the same items (Nation & Cocksey, 2009) should be included in longitudinal test batteries to test the separable effects of familiarity with the phonological form and semantic knowledge of a word.

In the pre-training for the orthographic learning study, only phonological aspects of the new words were trained, therefore any benefit on orthographic learning could not be due to semantic knowledge. Nonwords which received this pre-training were subsequently read more accurately. It is argued that when children make partial decoding attempts, which did indeed occur during orthographic learning, the taught phonological representation provides a pronunciation match and thus helps children to produce the correct answer. Critically the benefit from phonological pre-training was the same for children with Down syndrome and typically developing children when matched on target decoding. It is assumed that matching on this variable equates children on the production of partial decoding attempts and thus also equates the opportunity for pre-existing phonological representations to benefit reading. Therefore the magnitude of the effect of phonological pre-training on children's reading is dependent on their decoding ability.

The nature of semantics in the triangle model is unclear and has previously been represented as additional input from phonology (e.g. Plaut et al., 1996), although more realistic semantics have been implemented in later models (Harm & Seidenberg, 2004). However Taylor et al. (2011) suggested that lexical phonology could be incorporated within the triangle model, as semantic input could be re-conceptualised as item-specific phonological representations. Clearly further empirical work on the distinction between lexical phonology and semantics will also have implications for such theoretical claims.

5.2.4. Implications for Practice

Study 1 suggests that for children with Down syndrome, vocabulary may be more important for reading accuracy than phoneme awareness. However it is noteworthy that, although not significant, the relationship between reading accuracy and phoneme awareness was not non-existent. Coupled with the importance of phoneme awareness for reading unfamiliar words and some positive results from interventions that train phoneme awareness (Cologon et al., 2011; Goetz et al., 2008; Lemons & Fuchs, 2010a), it is suggested that phoneme awareness should be incorporated into reading instruction for children with Down syndrome. As vocabulary was a significant predictor of initial reading accuracy in the children with Down syndrome, this highlights the importance of oral language instruction and provides support for reading interventions which combine both oral language and phoneme awareness (e.g. Burgoyne et al., in press). Further research should aim to clarify whether children need to be familiarised with the phonological form of the word or also receive semantic instruction in interventions in order to benefit reading.

The relationship between vocabulary and reading accuracy in the children with Down syndrome in Study 1 and the benefit of phonological pre-training on orthographic learning in Study 3 are argued to reflect the resolution of partial decoding attempts using pre-existing phonological representations. Therefore typically developing children and children with Down syndrome should be introduced to the phonological form of an unfamiliar word before encountering it in print, for example when reading a new book.

5.3. The Effect of Reading Accuracy on Oral Language Development

5.3.1. Reading Accuracy as a Predictor of Oral Language Development

A range of oral language measures were included in Study 1 at each time-point, therefore the longitudinal effect of reading accuracy on phonology, grammar and vocabulary could be examined in children with Down syndrome and typically developing children. The suggestion that reading accuracy promotes oral language development in children with Down syndrome has been linked to their cognitive profile (Buckley & Bird, 1993), thus there is the implication that this relationship is stronger than in typical development.

Previous research has found an effect of reading accuracy on phoneme awareness in typical development (Morais et al., 1979), although it is clear that this relationship is not as strong as the causal effect of phoneme awareness on reading accuracy (Hulme et al., 2005). In Study 1, when controlling for age and nonverbal ability, reading accuracy was related to later phoneme awareness in the typically developing group but not for the children with Down syndrome. However the relationship was no longer significant in either group when the autoregressor was controlled for. Reading accuracy has also been found to predict nonword repetition in typically developing children (Castro-Caldas et al., 1998; Nation & Hulme, 2011) but this was not found for either group in the present study.

The effect of reading accuracy on the broader non-phonological oral language skills of vocabulary and grammar will now be considered. When age and nonverbal ability were controlled, there was no effect on later grammatical skills but in contrast earlier reading accuracy was related to later vocabulary in both groups. When the autoregressor was controlled for, only the correlation with picture naming in the typically developing group remained significant, although the correlation with receptive vocabulary in the children with Down syndrome was of a similar, moderate, strength. These results suggest that reading accuracy may indeed facilitate vocabulary development, but this is not unique to individuals with Down syndrome. With increasing reading practice, which could be reflected by reading accuracy level in the present study, children will encounter more words and therefore have more experience to expand their vocabulary. Furthermore if reading experience promotes segmental phonological representations (Ziegler et al., 2003), this may result in more precise phonological representations of known words. As breadth of vocabulary knowledge is thought to rely heavily on children's phonological representations of known words, this may in turn result in better performance on such tasks.

5.3.2. The Effect of Orthographic Support on Spoken Word Learning

Case studies of children with Down syndrome suggest an item-specific relationship between reading accuracy and vocabulary knowledge; specifically it has been proposed that presenting a word in print will benefit the production of the same word in speech (de Graaf, 1993; Duffen, 1976). In Study 2c, this claim was tested empirically. Seventeen children with

Down syndrome and 27 typically developing children were taught nonword names for unfamiliar pictures, half of which were taught with the orthography present. To ensure both groups had equal opportunity to benefit from the orthographic form, the typically developing children were selected to be of similar reading ability to the children with Down syndrome. The primary outcome measures were production measures: performance on production trials during learning and on a picture naming post-test. There were also two receptive mispronunciation tasks, which assessed children's ability to discriminate the taught nonwords from phonetically similar distracters.

The children's performance on the two production measures was improved by the provision of the orthographic form, as would be expected from similar studies with typically developing children (Ricketts et al., 2009; Rosenthal & Ehri, 2008). Importantly this effect was of a similar magnitude in both groups, suggesting that the benefit of orthography on phonological learning is not unique or special in individuals with Down syndrome and therefore is not due to their uneven cognitive profile.

A mechanism by which orthographic support may facilitate phonological learning is through grapheme-phoneme correspondences. Buckley (1995) argued that seeing the letters in a word enables children with Down syndrome to sound the word out thus improving production. In this manner, orthography could provide an opportunity for children to clarify or confirm the transient phonological representation. Rosenthal and Ehri (2009) present a similar argument, but suggest that the application of grapheme-phoneme knowledge is automatic. They argued that this process results in the spelling and pronunciation of the word becoming bonded together in memory, to be retrieved automatically, i.e. by word recognition. Whether grapheme-phoneme correspondences are activated implicitly or explicitly in children, the orthographic form of word would serve to promote the accuracy and strength of the phonological representation. In addition to improving the accuracy of the phonological representation, the orthographic form of the word is thought to provide an extra representation, and this strengthens the overall lexical representation (Perfetti & Hart, 2002; Ricketts et al. 2009).

Previous studies have produced mixed results as to whether children with Down syndrome show impairments in phonological learning (Jarrold et al., 2009 cf. Mosse & Jarrold, 2011). The results from the present study on the two production measures suggest that phonological learning is not impaired in children with Down syndrome. However the children with Down syndrome did show relative weaknesses on the receptive mispronunciation tasks. These results may have been due to difficulties in speech discrimination, memory or task understanding, and together with the previous mixed findings, the present results suggest a need for further replication and clarification.

The children with Down syndrome and typically developing children were matched on reading accuracy, and due to the status of reading as a strength in children with Down syndrome, they performed significantly poorer on a standardised test of vocabulary than the typically developing children. The discrepancy between the performance of the children with Down syndrome on the new word learning task and an existing vocabulary knowledge task could indicate a consolidation problem. Children with Down syndrome often have sleep disturbances (Miano et al., 2008; Shott et al., 2006) and the importance of sleep in the consolidation of word learning has been highlighted (Dumay & Gaskell, 2007). It is possible that a hypothesised problem in vocabulary consolidation could be connected with sleep abnormalities; this has also been suggested in other disorders such as autism (Norbury, Griffiths, & Nation, 2010). This is speculative and as such future research should first investigate vocabulary consolidation, by training children with Down syndrome and typically developing children to criterion and then testing retention. If children with Down syndrome show less retention over time, then the potential relationship with sleep disturbances could be explored.

5.3.3. The Effect of Reading Accuracy on Vocabulary Development: Item-Specific Versus General Underlying Mechanisms

The most convincing effect of reading accuracy on oral language in Study 1 was on vocabulary, as there was a moderate relationship in both groups even when controlling for the autoregressor. As noted above, the vocabulary tasks used in this study were heavily dependent on phonological representations of the word along with some semantic knowledge. Study 2 demonstrated that providing the orthographic form of a word helped children to learn and produce the correct phonological form, and this was to the same extent in both groups of children.

Reading practice may result in a detailed and more precise phonological system by promoting the segmental nature of words (Ziegler et al., 2003). If orthography has this off-line general effect on lexical restructuring then existing phonological representations may become better-specified. This argument has been applied to the task of nonword repetition (Nation & Hulme, 2011) but this may also feasibly have a beneficial effect on vocabulary tasks which require children to have accurate phonological representations of known words.

The relationship between reading accuracy and vocabulary may also be item-specific; as reading accuracy develops, children will encounter more words and therefore have more opportunity to acquire new vocabulary items through being exposed to their phonology and also their semantics through surrounding context. To test an item-specific relationship the reading accuracy and vocabulary tasks need to include the same words, which was not the case in the standardised measures used in Study 1. However Study 2 provided the orthography, phonology and a shallow semantic representation (picture) of all items and the presentation of orthography benefited phonological learning, which indeed suggests an item-specific effect. Provided the individual has sufficient letter-sound knowledge, the presentation of the graphemes within a word allows the target phonemes to be identified and this aids the formation of a well-specified phonological representation of the spoken word. Furthermore orthography provides extra information to and therefore strengthens the established lexical representation.

5.3.4. Implications for Practice

The results of Study 2, along with previous research with typically developing children may seem fairly intuitive but as both Rosenthal and Ehri (2008) and Ricketts et al. (2009) note, exposure to orthography is not promoted in established methods of oral vocabulary instruction. In an influential book by Beck et al. (2002), which outlines effective ways of teaching children new words, the orthographic form of the word is only shown to children after extensive instruction centred on the phonology and semantics of the word. The results of the present study suggest that the orthography of new words should be presented alongside phonological and semantic instruction. This would be simple for both parents and teachers to implement, by showing a flashcard of a word when introducing it in its spoken form. The continued presence of orthography after the first introduction of the spoken word may also be useful as phonological input can vary across individuals and contexts.

5.4. The Contributions of Reading Accuracy and Oral Language to Reading Comprehension

The educational attainment of children with Down syndrome has increased in recent years, most likely enhanced by the growing importance placed on early intervention programmes and mainstream schooling (Buckley et al., 2006; Chapman & Hesketh, 2000). As reading accuracy skills improve, this has enabled studies of reading comprehension in individuals with Down syndrome to be conducted. Reading comprehension appears to be an area of relative difficulty (e.g. Cardoso-Martins et al., 2009; Fletcher & Buckley, 2002; Nash & Heath, 2011; Verucci et al., 2006). Thus far there has only been one longitudinal study investigating reading comprehension and this did not include a control group, therefore Study 1 aimed to add to this body of research.

As the children with Down syndrome in Study 1 were matched to the typically developing children on reading accuracy at Time 1, they performed significantly more poorly on reading comprehension at this time-point, and indeed Time 2 and Time 3. The contribution of oral language to reading comprehension is proposed to vary as a function of reading accuracy skills (Vellutino et al., 2007). Indeed this has been argued to underlie the stronger relationship between oral language and reading comprehension reported in children with Down syndrome (Nash & Heath, 2011). If this is the case, when children are matched on reading accuracy, the contribution of oral language to reading comprehension should be equivalent in the two groups.

At Time 1, the children with Down syndrome and typically developing children were in the early stages of reading development and therefore as was predicted, in both groups reading accuracy significantly predicted concurrent reading comprehension whereas oral language did not. This is in line with similar findings with typically developing children (Ouellette & Beers, 2010; Vellutino et al., 2007) and extends this to children with Down syndrome. When children are beginning to learn to read, reading accuracy constrains the level of language they can access via written text and therefore plays a prominent role in determining performance on a reading comprehension task. Importantly this appears to be the same in both children with Down syndrome and typically developing children.

Longitudinally both reading accuracy and vocabulary predicted reading comprehension for the typically developing children. This is likely to be because their higher levels of reading accuracy now allowed them to access texts with more advanced language, and therefore oral language also becomes important for reading comprehension. For the children with Down syndrome, vocabulary predicted later reading comprehension whereas the variance predicted by reading accuracy was negligible. As this group of children were at a lower reading accuracy level at Time 3 than the typically developing children, reading accuracy was expected to be a *stronger* predictor than oral language. Therefore this was an unexpected finding, and a possible explanation relates to the reading comprehension task used. A greater proportion of scores for the children with Down syndrome than the typically developing children were based on a passage which required both reading and listening comprehension. Listening comprehension will clearly put more demands on oral language than reading accuracy and therefore likely contributed towards this pattern of results.

5.4.1. Implications for Practice

The importance of reading accuracy for reading comprehension seen in the concurrent correlations underscores how important effective reading accuracy instruction is for children with Down syndrome. Other factors that may be important for practitioners to consider are expressive language and verbal short-term memory limitations. Both MLU and verbal short-term memory were significantly correlated with reading comprehension for the children with Down syndrome only. The reading comprehension task used in the present study required children to answer open-ended questions about the text they have just read, which places clear demands on expressive language. If a child has severe expressive language difficulties then it may be advisable to use multiple-choice receptive questions for teachers to gauge a child's level of understanding. Reading stories and answering questions at the end of the text places high demands on verbal memory, and difficulty in this domain may limit children with

Down syndrome's understanding of the text. Shorter texts or separating the text into different sections with questions administered at interim points may result in lower memory demands. However it is important to remember that reading comprehension in everyday life does have high memory demands so perhaps altering texts in this way does not reflect real life.

5.5. Methodological Considerations

5.5.1. Sample Characteristics

The criteria for participation in all of the studies in this thesis required children with Down syndrome to use verbal communication and to be able to read according to parental report. Furthermore, most children were in mainstream school settings. Therefore they were a relatively high-achieving group of individuals and these findings may not be applicable to children with Down syndrome with lower cognitive abilities. Considering the example of Study 3, all children in this experiment were readers, and the results may be different when children are beginning to learn to read. In essence, it could be that the results of this study, particularly the ability to benefit from phonological pre-training, may be specific to those children with Down syndrome who are at the higher end of the spectrum of ability seen in this disorder.

School placement can affect the educational outcomes of children with Down syndrome; children in mainstream schools have been found to make more progress on language and literacy measures than children in special schools (Buckley et al., 2006), although there is typically a confound with IQ as children with lower IQs are more likely to attend special schools. In the group of children who participated in the three studies presented in this thesis, most attended mainstream schools although some were in special schools and some had mixed placements. The difference in school setting could be a confound, although the sample was not large enough to examine this systematically.

Much reference has been made to the commonly reported cognitive profile seen in individuals with Down syndrome. Indeed this has been the impetus for empirical work reported in this thesis and elsewhere. However there are individual differences and not all individuals with Down syndrome present with this profile. Indeed Tsao and Kindelberger (2009) found that only half of their sample of children with Down syndrome showed the expected pattern of strengths and weaknesses, with higher composite scores on nonverbal compared to verbal tasks in an IQ test battery. The remaining half of the sample was divided approximately equally into those who showed no difference between the two domains and those who had better verbal than nonverbal abilities. Furthermore within the nonverbal domain, there were discrepancies between performance on different subtests. This cautions against simplistic statements concerning the implied homogenous profile of strengths and weaknesses of children with Down syndrome.

An inherent problem with working with children from clinical populations is the issue of sample size. In Studies 1, 2 and 3, the number of children with Down syndrome participating was 21, 17 and 16, respectively. This is typical of those used in similar studies but is still relatively low, and this has several consequences for statistical analyses. Despite being of a moderate strength, some correlations were not significant, and some between-group differences in correlations were relatively large but not significant. The small sample size also restricted the number of predictor variables that were entered in path models for predicting longitudinal and concurrent outcomes in Study 1.

5.5.2. Matching Procedures

In Study 1, children with Down syndrome were pairwise matched to typically developing children on the basis of their reading accuracy, although extra children were recruited to the typically developing group to avoid potential problems with attrition. Reading accuracy was the main focus of this study and matching on this variable ensures that any differences between the groups were not due to their stage of reading accuracy development at Time 1. Due to the learning difficulties of the children with Down syndrome they made slower progress than the typically developing children and therefore were at a lower stage of reading accuracy ability at Time 3. It is important to bear in mind that this may affect any differences between the two groups regarding the longitudinal predictors of reading accuracy. It should also be highlighted that matching on reading accuracy does not necessarily mean that the children with Down syndrome and the typically developing children are using the same strategies or routes to reading (Roch & Jarrold, 2008)

Due to the strengths and weaknesses associated with Down syndrome (but see Tsao and Kindelberger (2009)) matching on one task leads to differences from typically developing children on other tasks. In the context of Study 1, matching on reading accuracy led to lower levels of phonological awareness in the children with Down syndrome. Therefore it is difficult to select tasks that are appropriate for children with Down syndrome and typically developing children, for example the phonological awareness task was relatively simple to avoid floor effects for the children with Down syndrome but this resulted in ceiling effects for the typically developing children.

The method by which the groups were matched also warrants discussion. In Study 1, pairwise matching was used, i.e. typically developing children were individually matched to the children with Down syndrome. This was done to ensure the groups were at the same level of reading accuracy and not only would the groups be matched on the mean reading score but the range of scores would also be equivalent in the two groups (Facon et al., 2011). However there is some controversy regarding the use of appropriate control groups in studies with children with developmental disabilities (Jarrold & Brock, 2004; Thomas et al., 2009). A larger normative sample that is not explicitly matched to the atypical group is an approach advocated by Jarrold and Brock to avoid problems with generalisability and interpretation that can occur when groups are matched on a specific variable. This method was used in the word learning studies, where slightly larger groups of typically developing children were recruited from year groups thought to be at a similar reading level as the children with Down syndrome. Recruiting a larger typically developing group also meant that there was the possibility of identifying typically developing subgroups matched on different variables to the children with Down syndrome. Indeed in the orthographic training study, a subgroup of typically developing children were matched to the children with Down syndrome on target decoding, which revealed that children with Down syndrome showed orthographic learning and a benefit from phonology in line with their decoding ability.

5.6. Summary and Conclusions

This thesis investigated the reciprocal relationship between reading and oral language skills in children with Down syndrome. This has typically been examined by looking at the level of, and correlations between acquired skills concurrently and longitudinally. Dynamic training studies offer a complementary perspective by focussing on the processes underlying skill acquisition, although this method has not yet been used extensively with children with Down syndrome.

There were three overarching areas of interest that the empirical work in this thesis aimed to address: the effect of phonological and non-phonological skills on reading accuracy, the effect of reading accuracy on oral language development and the relative contributions of reading accuracy and oral language to reading comprehension. Study 1 was conducted to address all three of these areas, by the longitudinal assessment of reading accuracy, reading comprehension and oral language in children with Down syndrome and typically developing children matched on reading accuracy. Study 2, a spoken word learning study, examined the effect of orthographic support on phonological learning, in order to speak to the proposed benefit on oral language development from reading. Study 3, an orthographic learning study, investigated the effect of phonology on reading accuracy, by examining the effect of phonological pre-training on learning to read new words.

In line with previous research, the effect of phoneme awareness on reading accuracy in Study 1 in children with Down syndrome was weaker than would be expected from studies with typically developing children. In contrast, vocabulary was a significant predictor of reading accuracy. Due to the high levels of longitudinal stability, these results were based on concurrent relationships. In this thesis it has been argued that performance on the vocabulary tasks heavily reflects lexical phonology, and it is this which underlies the relationship between vocabulary and reading accuracy in the children with Down syndrome by resolving partial decoding attempts.

Study 3 demonstrated that children with Down syndrome show orthographic learning below the level expected from their reading accuracy. However this performance was in line with decoding ability; this is likely to be because phonological recoding ability was equated and this is what primarily underlies orthographic learning according to the self-teaching hypothesis. When compared to the full sample of typically developing children matched on reading accuracy, the children with Down syndrome appeared to benefit more from phonological pre-training. However, when a typically developing subgroup was matched on target decoding, the two groups benefited to the same extent. This suggests that the extent of the benefit provided by phonological pre-training is dependent on decoding ability, presumably because children's decoding accuracy determines the number of partial decoding attempts made and also therefore the number of opportunities for pre-existing phonological representations to benefit reading. This argument can also be applied to the results from the longitudinal study; as the children with Down syndrome had poorer decoding ability, there were more opportunities for phonological representations to supplement partial decoding attempts. A logical extension from this is that if children with Down syndrome and typically developing children were matched on decoding ability in a correlational study, the relationship between this skill and vocabulary should be similar.

It has been proposed that learning to read benefits the oral language of children with Down syndrome due to their cognitive profile (Buckley & Bird, 1993), which implies a stronger relationship than in typically developing children. This was not supported by Study 1 nor Study 2. Study 1 found relatively little evidence for the effect of reading accuracy on the development of phonological skills although there was a moderate effect on vocabulary, but importantly this was not greater for the children with Down syndrome. It is argued that this might occur because reading experience promotes the re-organisation of existing phonological representations into more segmental forms and exposes children to new words. Study 2 found that presenting children with the orthographic form of a word helps them to produce its spoken form, and this benefit was to the same extent in children with Down syndrome and typically developing children. Orthographic support is suggested to both improve the quality of the phonological representation and strengthen the overall lexical representation. The combined results from Studies 1 and 3 provide clear evidence that the effect of reading on oral language is not unique or greater for children with Down syndrome, but rather that there is a benefit for this population of children and typically developing children.

The combined evidence from the training studies and the longitudinal study strongly suggests the presence of a mutually beneficial relationship between written and spoken language. Specifically the present results suggest that when teaching the phonological form of a word, knowledge of the orthographic form is beneficial and vice-versa. Therefore this suggests high-quality information in multiple modalities leads to enhanced lexical representations in line with the lexical quality hypothesis (Perfetti & Hart, 2002).

The simple view of reading (Gough & Tunmer, 1986) was also evaluated in children with Down syndrome relative to typically developing children in Study 1. The results suggested that the relative contributions of reading accuracy and oral language to concurrent reading comprehension are dependent on reading accuracy levels, rather than group membership. However the longitudinal results were mixed and there remains a need for further longitudinal studies of reading comprehension in children with Down syndrome.

In summary, when children with Down syndrome are matched to the appropriate ability to their typically developing control group, the interaction between reading and oral language skills has more similarities than differences. Coupled with findings from previous research, phoneme awareness appears to play a weaker, yet still important, role in the development of reading accuracy but the effect of pre-existing phonological representations on reading accuracy is argued to be similar when children are equally able to decode unknown words. The effect of reading accuracy on spoken language is similar to typical development when children are matched for their ability to access the written form. The findings of this thesis highlight the importance of comparing results to control groups matched on the appropriate variables and demonstrate both the feasibility and utility of conducting carefully-controlled learning studies with this population of children.

Appendix 1

Reliability Estimates for Standardised Tests

Test name	Reliability estimate	Measure of reliability reported
WPPSI-III[UK] Matrices	.90	Spearman-Brown[a]
WASI Matrices	.92	Spearman-Brown
YARC Letter-Sound Knowledge	.98	Cronbach's alpha[b]
YARC Early Word Reading	.98	Cronbach's alpha
YARC Single Word Reading	.98	Cronbach's alpha
YARC Passage Reading Form A Reception passage	.77	Cronbach's alpha
YARC Passage Reading Form A Level 1	.64	Cronbach's alpha
YARC Passage Reading Form A Level 2	.62	Cronbach's alpha
YARC Passage Reading Form A Level 3	.48	Cronbach's alpha
YARC Passage Reading Form A Level 4	.67	Cronbach's alpha
YARC Passage Reading Form A Level 5	.57	Cronbach's alpha
YARC Passage Reading Form A Level 6	.66	Cronbach's alpha
GNWRT	.96	Cronbach's alpha
YARC Sound Deletion	.93	Cronbach's alpha
ERB PSRep	.89	Cronbach's alpha
WMTB-C Digit Recall	.82[c]	Pearson's r[d]
WMTB-C Block Recall	.53[c]	Pearson's r
WMTB-C Word Recall	.72[c]	Pearson's r
WPPSI-III[UK] Picture Naming	.88	Spearman-Brown
BPVS	.93	Cronbach's alpha
	.86	Spearman-Brown
CELF 4[UK] Sentence Structure	.70	Cronbach's alpha
	.80	Pearson's r
	.71	Spearman-Brown
CELF-Preschool 2[UK] Sentence Structure	.78	Cronbach's alpha
	.78	Pearson's r
	.80	Spearman-Brown

[a] a measure of split-half reliability
[b] a measure of internal consistency
[c] mean created from estimates reported for Years 1 and 2 and for Year 5 and 6
[d] a measure of test-retest reliability

Appendix 2

Items for the Alliteration Matching Task (Taken From Carroll, 2004)

Trial	Target item	Correct answer	Distracter matched on global similarity
Practice	Hit	Hose	Rake
Practice	Pot	Peach	Duck
Test	Nail	Nose	Bed
Test	Tap	Tin	Leg
Test	Pig	Pool	Beak
Test	Map	Moon	Net
Test	Beak	Bowl	Shed
Test	Feet	Fan	Sick
Test	Sock	Sun	Fat
Test	Chin	Chop	Shell
Test	Cage	Coat	Head
Test	Bin	Boat	Game

Appendix 3

Items for the Phoneme Deletion Task (Taken from McDougall et al., 1994)

Trial	Item (phoneme to be deleted)
Practice	(c)at
Test	(b)ice
Test	toa(b)
Test	(b)arch
Test	tea(p)
Test	(k)elm
Test	bloo(t)
Test	jar(l)
Test	(g)lamp
Test	(b)rock
Test	ma(c)t
Test	s(t)ip
Test	hi(f)t
Test	star(p)
Test	c(r)oal
Test	(f)rip
Test	hil(f)
Test	cro(t)s
Test	c(l)art
Test	bir(l)d
Test	fors(k)
Test	s(p)low
Test	(s)trail
Test	(b)eel
Test	cloo(f)

Appendix 4

Narrative Content Marking Guidelines

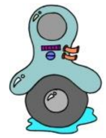
	3 points	2 points	1 point	0 points
Unit 1		Snow (walking in the) snow	Bad weather Winter	
Unit 2	*Children + walking + school* Children walking to school	*2 of children, walking, school* (People are) walking to school *(i.e. without 'children')* Girl OR boy walking to school	*1 of children, walking, school* People walking/they walk *(i.e. without 'school' and 'children')* Arrive at school (*i.e. without 'walking' and 'children')*	Saw a school
Unit 3		Sign that said school is closed	School is closed No school	
Unit 4	*Who(children) and what* The other children are playing in the snow The other children are making snowmen	*Without who or without detail of snow* (they are) making snowmen (they are) throwing snowballs The other children/people are playing *(no subject)* playing in the snow	There are children in the playground (There is a) snowman	
Unit 5		The boy and girl play by the frozen lake/ice/in the snow	The boy and girl go and play The boy/girl go to a frozen lake/ice Boy OR girl play by the lake/in the snow The boy and girl make snowmen *(i.e. implication of playing in the snow)* *(no subject)* playing in the snow	The boy and girl go to the park/home

Unit 6	There is a 'keep-off' sign	The lake is dangerous The girl shouldn't have gone on the lake It says 'keep off' There was a sign	
Unit 7	*Girl and ice* The girl goes onto the lake/ice The girl plays on the lake/ice The girl goes (one person is) ice-skating	The girl plays Ice-skating	The boy goes onto the lake The girl went on the ice rink They go on the ice Girl skiing
Unit 8	*Falling and ice breaking* The ice cracks and the girl falls in	She went into the water Falling Cracking The girl cracks the ice It cracked	There was a cracking noise Falling ON the ice The girl got wet Crash Girl fell over
Unit 9	The boy helps her		
Unit 10	A passer-by comes over to help A passer-by pulls her out of the lake	A passer-by comes over Someone *(doesn't matter who)* pulled her out	
Unit 11	(The girl is) in bed	The girl has to go home (The girl goes to) sleep	They go to sleep Bed(time)
Unit 12	The girl is unwell The girl had a cold/temperature Poorly	The girl is hurt	Man poorly Girl broke her (any body part)
Unit 13	The doctor is there The doctor comes to see the girl	(She went to) hospital/the doctors	Her mum rung the doctors
Unit 14	The mum and brother are worried/upset	The mum/boy is worried/upset	

Appendix 5

Nonwords and Pictures Used in Study 2

	Target nonword	Phonetic transcription	'Alien' spelling	Picture paired with the target nonword	Distracter in the multiple-choice mispronunciation task (initial phoneme changed)	Distracter in the multiple-choice mispronunciation task (final phoneme changed)	Nonword presented in the forced-choice mispronunciation task
Word group A	vum	/vʌm/	ϐΦΩ		zum	vun	fum
	sav	/sæv/	θφΛ		zav	saf	saz
	tid	/tɪd/	πΨϑ		pid	tib	tid
	pon	/pɒn/	ϨϪφ		bon	pom	pon

	mep	/mep/	Ψφƛ		nep	meb	mep
Word group B	pag	/pœg/	ƛϐΘ		kag	pab	pag
	deg	/deg/	ΘΦΨ		teg	deb	geg
	zot	/zɒt/	Λζθ		vot	zop	zod
	yub	/jʌb/	ϑϕ⋟		wub	yud	yug
	miv	/mɪv/	Ω⋟δ		niv	miz	miv

Appendix 6

Nonwords Used in Study 3

	Target nonwords	Phonetic transcription	Phonological distracter with one phoneme change	Phonological distracter with two phoneme changes	Homophone distracter	Visual distracter
Word group A	keve	/kiːv/	geve	geze	keav	kewe
	nirp	/nɜːp/	nirt	mirt	nurp	nirg
	vabe	/vaɪb/	vame	zame	vaib	vade
	kewf	/kjuːf/	pewf	pewv	kufe	kewk
	roak	/rəʊk/	loak	loag	roke	roaf
	gorn	/g ɔːn/	gord	dord	gawn	gorm
Word group B	rorb	/rɔːb/	lorb	lorp	rawb	rorp
	nawg	/nɔːg/	nawp	mawp	norg	nawy
	zale	/zaɪl/	zade	vade	zail	zafe
	lirg	/lɜːg/	lird	dird	lurg	lirp
	joat	/jəʊt/	choat	choap	jote	joaf
	neam	/niːm/	zeam	zean	neme	nean

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