

READING SKILLS IN DOWN SYNDROME:  
AN EXAMINATION OF ORTHOGRAPHIC KNOWLEDGE

by

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## ABSTRACT

The primary purpose of the present study was to examine orthographic knowledge in a sample of individuals with Down syndrome (DS) in comparison to typically developing (TD) children matched on word identification ability. Phonological recoding was also compared across groups. A secondary purpose of the study was to estimate literacy rates in DS using parent reports. Results of the parent study revealed that most individuals with DS over the age of 5 years can read words. Results of the child study revealed that individuals with DS performed worse than TD controls on a measure of phonological recoding, but similarly on two measures of orthographic knowledge. Interestingly, the group with DS performed worse on a third measure of orthographic knowledge. The first two orthographic tasks both included real words as stimuli; the third orthographic task used letter patterns, but did not include real words. These results suggest that individuals with DS may have a relative strength in word-specific orthographic knowledge but not in general orthographic knowledge.

## DEDICATION

This dissertation is dedicated to mothers. To the mothers of my participants, who never stop believing in or fighting for their children, I am continually amazed and touched by your dedication and love. To my own mother, who is my rock and inspiration, thank you for telling me I could do anything I set my mind to, for believing it, and for helping me believe it. My success is your success. Love you.

## LIST OF ABBREVIATIONS AND SYMBOLS

Cohen's $d$	Measure of effect size
$F$	Fisher's $F$ ratio: a ratio of two variances
$M$	Mean
$Md$	Median
$p$	Probability associated with the occurrence under the null hypothesis of a value as extreme as or more extreme than the observed value
$r$	Pearson product-moment correlation
$t$	Computed value of $t$ test
$n$	Sample size for group
$SD$	Standard deviation
$U$	Computed value of Mann-Whitney U test
$z$	$z$ value
$>$	Greater than
$<$	Less than
$=$	Equal to
ADHD	Attention deficit hyperactivity disorder
ANOVA	Analysis of variance
CASL	Comprehensive Assessment of Spoken Language
CELF	Clinical Evaluation of Language Fundamentals – Fourth Edition
DS	Down syndrome

EVT-2	Expressive Vocabulary Test – Second Edition
EWR	Exception Word Reading
HLE	Home literacy environment
ID	Intellectual disability
IQ	Intelligence quotient
KBIT-2	Kaufman Brief Intelligence Test – Second Edition
MANOVA	Multivariate analysis of variance
OA	Orthographic Awareness
OC	Orthographic Choice
PPVT	Peabody Picture Vocabulary Test – Fourth Edition
PR	Phonological recoding
RA	Reading ability
RAN	Rapid automatized naming
RM(C)	Reading motivation as reported by the child
RM(P)	Reading motivation as reported by the parent
TD	Typically developing
UAIDPR	University of Alabama Intellectual Disabilities Participant Registry
WID	Word identification
WISC-IV	Weschler Intelligence Scale for Children – Fourth Edition
WRMT-III	Woodcock Reading Mastery Test – Third Edition

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## CONTENTS

ABSTRACT .....	ii
LIST OF ABBREVIATIONS AND SYMBOLS .....	iii
ACKNOWLEDGEMENTS .....	v
LIST OF TABLES .....	ix
LIST OF FIGURES .....	x
INTRODUCTION .....	1
METHODOLOGY .....	31
RESULTS .....	42
DISCUSSION .....	56
REFERENCES .....	69
APPENDICES .....	83

## LIST OF TABLES

1. Children’s Demographics .....	31
2. Means and Standard Deviations of Sample Descriptives .....	34
3. Parent Report of Child’s Reading Abilities .....	43
4. Number of Children’s Books in the Home by Child’s Age .....	44
5. Time Parent Reads to/with Child by Child’s Age and Age Parent Began Reading to Child .....	44
6. Child’s Motivation to Read .....	45
7. Correlations between Age, Reading Ability, Home Literacy Environment and Reading Motivation .....	46
8. Means, Medians and Standard Deviations of Child Study Variables .....	50
9. Means and Standard Deviations of Orthographic and Phonological Task Z-scores .....	51
10. Correlations between Receptive Vocabulary, IQ, and Reading Measures .....	54
11. Correlations between Parent Survey and Comparative Child Studies .....	55

## LIST OF FIGURES

1. Simple View of Reading .....	3
2. Recognition vs. Production Measures of Orthographic Knowledge .....	51
3. Phonological Recoding vs. Orthographic Knowledge .....	53

## INTRODUCTION

It was once believed that individuals with intellectual disabilities (ID) could not learn to read. Today, while the exact literacy rates in ID are not known, research has indicated that individuals with ID can, in fact, learn to read. Further, research has begun to examine strengths and weaknesses in various etiologies of ID, including Down syndrome (DS). Individuals with DS show evidence of strong word identification skills relative to mental age, but poor phonological recoding skills relative to word identification ability. Research on orthographic processing in DS is very scarce and has yet to be fully explored. The purpose of the present study is to examine orthographic processing in a sample of individuals with DS to better understand reading abilities in this population.

### **Down Syndrome**

Down syndrome is the leading genetic cause of ID. It is believed to occur in approximately one in every 733 live births (Canfield, et al., 2006). It is caused by having a complete or partial triplication of chromosome 21. Down syndrome is associated not only with mild to severe intellectual disability, but also with a specific phenotype and cognitive profile.

Physically, individuals with DS tend to be of short stature, have short necks, broad, short hands with a single palmar fold and specific facial characteristics (Korenberg et al., 1994). These facial characteristics include round, flat faces, flat nasal bridges, enlarged tongues, small chins, small oral cavities, epicanthic folds and brushfield spots on their eyes. Additionally, DS is associated with several health concerns, such as increased risks of congenital heart disease (Dykens, Hodapp, & Finucane, 2000), thyroid dysfunction (Siegfried, Pueschel, & Pezzullo,

1985), being overweight and/or obese (Bell & Bhate, 1992), and developing Alzheimer's disease (Janicki & Dalton, 2000; Lai & Williams, 1989). Hearing impairments are also prominent in DS (Meuwese-Jongejueugd, et al., 2006).

Cognitively, individuals with DS have known patterns of impairment including poorer performances in verbal domains and relatively stronger performances in visuospatial domains (Conners, Moore, Loveall, & Merrill, 2011). Particularly striking in the DS cognitive profile are poor speech and language abilities, even beyond what would be expected based on IQ (Abbeduto et al., 2003; Fidler, Most, & Guiberson, 2005; Kernan & Sabsay, 1996; Vicari, Casselli, Gagliardi, Tonucci & Volterra, 2002). While receptive vocabulary has been found to be on par with nonverbal ability (Abbeduto et al., 2003), expressive vocabulary is below developmental level for most individuals with DS (Chapman, Seung, Schwartz, & Kay-Raining Bird, 1998). Further, on measures of syntax, individuals with DS perform below their developmental level in both expressive and receptive skills (Abbeduto et al., 2003; Kernan & Sabsay, 1996; Vicari et al. 2002). A meta-analysis by Naess, Halaas Lyster, Hulme, and Melby-Lervag (2011) reported significant differences between individuals with DS and typically developing (TD) controls matched on nonverbal mental age in receptive vocabulary, expressive vocabulary and receptive grammar. Working memory, the system that deals with current information, whether newly learned or pulled from long-term memory, is also impaired in DS (Jarrold & Baddeley, 2001; Marcell & Weeks, 1988; McDade & Adler, 1980), beyond even what would be expected for mental age (Kay-Raining Bird & Chapman, 1994; Mackenzie & Hulme, 1987) and IQ (Fidler et al., 2005; Marcell, Harvey & Cothran, 1988; Marcell, Ridgeway, Sewell & Whelan, 1995; Marcell & Weeks, 1988; McDade & Adler, 1980). Naess and colleagues' (2011) meta-analysis

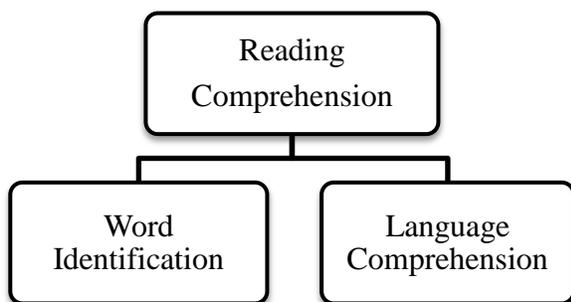
also examined verbal short term memory and found deficits for those with DS in comparison to TD participants matched on nonverbal mental age.

## Reading Skills

The ultimate goal of reading is comprehension (Catts & Kamhi, 1999), a higher-level process that involves understanding text. Reading is a complex skill, and comprehension builds off of lower-level skills. According to the Simple View of Reading (Gough and Tunmer, 1986; see Figure 1), reading comprehension has two main components, word identification and language comprehension. Word identification allows one to recognize words when reading, while language comprehension allows one to understand the meaning behind those words. The relative contribution and importance of these components changes across development (Gough, Hoover, & Peterson, 1996; Vellutino, Tunmer, Jaccard, & Chen, 2007). During earlier stages of reading development, word identification is the primary focus. It is not until an individual can accurately identify words that language comprehension becomes the primary focus. Because the present study is on orthographic processing in DS, the following review will cover several reading skills but focus primarily on orthographic processing.

Figure 1

### *Simple View of Reading*



**Theories of word identification.** Word identification refers to the pronunciation of a word, not necessarily understanding of the word's meaning. There are two major camps of theories on word identification: dual-route and connectionist. Dual-route theories of word identification (see Coltheart, 1978; 1980; 1996; 2000; Coltheart, Curtis, Atkins & Haller, 1993; Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001; Forster & Chambers, 1973) are step-by-step processing models that focus on the act of reading aloud –the pronunciation of a written word. These models include two routes: lexical and nonlexical. The term *lexicon* refers to stored information about the meaning and pronunciations of words, often coined a mental dictionary for words. The lexical route involves an individual looking up a familiar word stored in his/her lexicon. Each word in the lexicon has a meaning, and when individuals access words they also access the meanings of those words. In English, letters and graphemes (the smallest units of written words; letters or letter sequences that are used to represent phonemes) are used to visually represent phonemes, or speech sounds. If a word is new or unfamiliar, the nonlexical route is utilized. The nonlexical route is a rule-based route which involves translating graphemes into phonemes, or in other words, using the orthographic (visual) structure of a word to phonologically recode (verbalize) it. The sequence of graphemes is blended together to form the pronunciation of a word. Once unfamiliar words become familiar, they are then stored in the lexicon, and an individual can access them via the lexical route while reading.

The majority of English words, approximately 80%, can be considered “regular” in terms of the grapheme-phoneme, or letter-sound, relationship (Woodcock, 1987). This means that most words in the English language can be sounded out correctly based on their grapheme representations. For example, if a child encountered an unfamiliar word while reading, such as “red”, he or she could still read it by matching the graphemes to their corresponding

sounds/phonemes. However, the remaining 20% of words are irregular, meaning that they cannot be correctly sounded out based on their grapheme representations. These are known as “exception words”. *Salmon*, *love*, and *eye* are all examples of exception words. According to dual-route theories, exception words cannot be sounded out via the nonlexical route so must be memorized and stored in the lexicon.

Connectionist theories (see Seidenberg, 2005; Seidenberg & McClelland, 1989; Plaut & McClelland, 2000; Plaut, McClelland, Seidenberg, & Patterson, 1996) also note the importance of both phonological and orthographic aspects of word identification. However, these theories are developmental and focus on how individuals master print-to-sound pronunciations. According to connectionist theories, the ability to recognize and pronounce words involves statistical learning. These models are designed as networks composed of neuron-like groups for spellings (orthography) and pronunciations (phonology). Each of these groups contains a large set of patterns. Instead of the exact grapheme-phoneme matches used in dual-route models, spelling-sound correspondences are represented on a continuum, the strength of which is determined by its connection between the spelling and pronunciation groups. In this model, exception words, while less common, do not have to be memorized. Instead, they can be learned using the same processes as regular words. This makes sense, as even exception words have some “regular” aspects to them. For example, Seidenberg (2005) notes that while the word *pint* is not regular, it does share some structure with regular words like *pant* and *pine*, and that learning the pronunciations of each can influence the learning of the others. The *p*'s in all three words, even the irregular *pint*, receive the same, regular pronunciation.

The Triangle Model by Plaut, McClelland, Seidenberg, and Patterson (1996; see also Seidenberg & McClelland, 1989) is a great example of a connectionist model. According to the

triangle model, there are three groups or “layers” that encode different types of information: phonology, orthography and semantics. These groups are typically drawn in a triangle pattern with semantics at the top. The phonology group is responsible for pronunciation and sound, the orthography group for spelling, and the semantics group for meaning. Processing involves an input group (i.e. orthography) activating an output unit (i.e. phonology). These connections establish important patterns of activity, such as a phonological pathway between orthographic and phonological groups and a semantic pathway between orthographic and semantic groups. For beginner readers, the phonological pathway may be most relevant, creating links from orthography (a word’s spelling) to phonology (the word’s pronunciation), which can later link to semantics (the word’s meaning). For these readers, both orthography and phonology work together to determine semantics. For experienced readers, the semantic pathway may be more relevant, linking orthography (a written sentence) to semantics (the meaning of the sentence). These three groups are also connected and routed through smaller units, which allow for deeper processing. For example, these smaller units may account for things like the order of letters in a word (i.e., on vs. no), in which context a word commonly occurs, and they may even allow for a feedback loop to the input group.

The dual-route models (e.g., Coltheart, 2000) offer a plausible explanation of how individuals with DS might be able to capitalize on stronger visual skills to counteract poorer verbal skills. According to this model, individuals with DS may be relatively stronger at reading whole words using the lexical route and much weaker at sounding out new or unfamiliar words via the nonlexical route. However, dual-route theories are not developmental, and therefore do not explain how phonological skills are developed (Hulme, Goetz, Brigstocke, Nash, Lervag, & Snowling, 2012). In contrast, connectionist models (e.g., Plaut et al., 1996) can be used to

explain how reading development can be arrested in DS based on the DS cognitive profile (see Bishop & Snowling, 2004 for similar work with children with oral language impairments). Poor verbal ability may lead to problems developing a strong phonological pathway, while stronger visual skills could lead to a relatively stronger orthographic and semantic pathway.

Several broader theories have focused on stages or phases of reading development (Chall, 1983; Ehri, 1998; 1999; 2002; Frith, 1985; Gough & Hillinger, 1980; Marsh, Friedman, Welsch, & Desberg, 1981; Mason, 1980; Seymour & Duncan, 2001; Stuart & Coltheart, 1988). Each of these theories portrays the growth of different processes and skills that emerge throughout reading development, from pre-reading to early reading to decoding/recoding and finally to fluent reading. Frith's (1985) theory consists of three stages: logographic, alphabetic and orthographic. In the logographic stage, links are made between the visual features of a word and its pronunciation. This stage is typically defined by a "sight vocabulary", or recognizing full words, such as colors or one's name. In this stage, children can read words they have been taught but cannot read unfamiliar words. In the alphabetic stage, links are made between letters and sounds whenever a new word is encountered. It is during this stage that children learn that language is represented by symbols and will sound out words based on their written form. In the orthographic stage, the ability to recognize words by their visual form emerges. These readers no longer have to sound out words phoneme by phoneme but can read words based on their visual form. It is in this orthographic stage that readers become quick and fluent. Further, this stage is important in that it allows readers to recognize exception words without the need to sound them out by phonemes, which could lead to an inaccurate production. Despite their "stages", theories of reading development are not so strict as to claim that one stage must be completely mastered before moving onto the next stage (Ehri, 2010). Instead, these theories

recognize that while skills related to reading often develop in succession, earlier and later stages often overlap in development.

**Phonological processing.** Both major theories of word identification (dual-route and connectionist) identify two important aspects of word identification: phonological processing and orthographic processing, or in other words, verbal (phonological) and visual (orthographic) aspects utilized in word reading. Both of these subskills account for significant, independent variance in word identification (Holland, McIntosh & Huffman, 2004) and will be examined in the present study. Phonological recoding is the process of sounding out, or translating a printed word into a speech-based form. Because it is difficult to know and control for which real words are unfamiliar to participants in reading studies, phonological recoding is typically measured by asking participants to sound out nonwords (e.g., *straced*). Since participants have never been exposed to them, “regular” nonwords measure a participant’s ability to phonologically recode.

Phonological recoding is just one of several phonological processing skills. Phonological recoding itself has two important subskills: phonological awareness and phonological memory (see Lonigan, Anthony, Phillips, Purpura, Wilson & McQueen, 2009). Phonological awareness refers to an awareness and sensitivity to the sounds of language, such as counting the number of syllables in a word. It also involves the manipulation of speech sounds, such as rhyming words. Phonological memory refers to the storage of speech sounds in working memory. Together, these three skills make up what is commonly referred to as “phonological processing” ability.

Research has indicated a strong link between phonological processing and word identification. Not only does phonological processing explain significant, unique variance in current word identification ability (Connors, Loveall, Moore, Hume, & Maddox, 2011; Cutting & Denckla, 2001; Kirby, Parrila, & Pfeiffer, 2003), it also predicts future word identification

ability (Kirby et al., 2003; MacDonald & Cornwall, 1995; Mann, 1993; Parrila, Kirby, & McQuarrie, 2004; Wagner, Torgesen, & Rashotte, 1994; Wagner et al., 1997) and future reading comprehension (Kirby et al., 2003). In a five-year longitudinal study Kirby and colleagues (2003) found that phonological awareness ability in Kindergarten explained 22% of concurrent word identification ability. Further, phonological awareness ability in Kindergarten predicted between 31-41% of word identification ability in Grades 1-5, after controlling for general mental ability and prior achievement. Another study by MacDonald and Cornwall (1995) found that phonological ability in Kindergarten (ages 5-6 years) predicted word identification 11 years later at 16 to 17 years of age. As Parrila and colleagues (2004) stated, “In sum, existing research suggests a developmental progression from phonological processing skills to word reading, and further to passage comprehension” (p. 4).

It is important to take a moment here to introduce another reading skill called rapid automatized naming (RAN), which has also become of interest to reading researchers. RAN refers to the ability to quickly name visual symbols such as letters, digits, colors or objects (Denckla & Rudel, 1974) and is predictive of reading ability, including word identification, reading comprehension and phonological recoding (Ackerman & Dykman, 1993; Compton, 2000; Manis, Seidenberg, & Doi, 1999; Schatsneider, Fletcher, Francis, Carlson, & Foorman, 2004). Although RAN is an important reading skill, there is debate within the literature as to where to place RAN among the other predictors of reading. Some research suggests that RAN is a third, separate, unique subskill of word identification (Cutting & Denckla, 2001; Manis, Doi, & Bhadha, 2000; Wolf, 1997; Wolf & Bowers, 1999), while others argue that it is a subskill of phonological processing (Torgesen, Wagner, Rashotte, Burgess, & Hecht, 1997; Wagner, Torgesen, Laughon, Simmons, & Rashotte, 1993; Wagner et al., 1994). While that debate is

outside the realm of this study, the reader should note that RAN is an important reading skill and will be mentioned throughout the document. RAN will not be examined in the present study as articulation issues commonly associated with DS (see Roberts et al., 2005; Roberts, Price, & Malkin, 2007) make these tasks complicated beyond the scope of the study.

Because of its central role and strong relationship to word identification, research in the TD literature has primarily focused on phonological processes. In fact, Snowling and Hulme (2010) report, “It appears that a consensus has been reached: phonological coding is central to word recognition” (p. 5). Yet some researchers have argued that while phonological processes are critical, they are not sufficient for the development of word identification (Juel, Griffith, & Gough, 1986; Reitsma, 1989; Tunmer & Nesdale, 1985). In fact, research on reading disabilities suggests that some children with adequate phonological processing skills still lag behind in word identification (Badian, 1996; Bowers & Swanson, 1991; Lovett, 1987; McBride-Chang & Manis, 1996; Wolf, 1991; Wolf & Bowers, 1999). While the research obviously demonstrates that phonological processing is important to word identification, it seems phonological processing is not the only important contributor to word identification. Research has therefore begun to examine the other subskill of word identification: orthographic processing.

**Orthographic processing.** Orthographic processing is the visual aspect of reading. It refers to the abilities used to visually recognize letter combinations found in words, to recognize what words look like, to distinguish correct spellings of words and to learn new word spellings. While much less studied than phonological processing, orthographic processing is an important subskill of word identification. One study by Holland et al. (2004) using structural equation modeling found that orthographic processing, not phonological processing, was the best predictor of word identification. Other studies have found that orthographic processing accounts

for significant variance in word identification even after statistically controlling for phonological recoding (Barker, Torgesen, & Wagner, 1992; Berninger, Cartwright, Yates, Swanson, & Abbott, 1994; Cunningham, Perry, & Stanovich, 2001).

There are two major areas of research on orthographic processing – studies of cumulative orthographic knowledge and studies of current orthographic learning. Cumulative orthographic knowledge reflects prior experience with print, which has accumulated over an individual’s years of reading. This involves knowledge about the positions and frequencies of where letters are typically found in words in one’s language that is assimilated throughout many years of reading. Current orthographic learning reflects current learning of a new set of orthographic features, typically from nonwords or unfamiliar new words. The present study will focus on cumulative orthographic knowledge in DS.

Cumulative orthographic knowledge is usually measured using letter string/orthographic awareness (see Siegel, Share & Geva, 1995), orthographic choice (see Olson, Forsberg, Wise & Rack 1994b), homophone choice (see Olson, Forsberg & Wise, 1994a) and/or exception word reading tasks (see Adams and Huggins, 1985). Letter string/orthographic awareness tasks visually present participants with two nonwords and ask which looks more like a real word (e.g., *thomer* vs. *thmoer*). Because these letter strings are not real words, participants must rely on their accumulated knowledge of the positional frequencies and combinations of letter patterns acquired throughout their reading experiences. Orthographic choice tasks also visually present participants with two options: one real word and one pseudohomophone, and participants are asked to select the real word (e.g., *room* vs. *rume*). Homophone choice tasks present participants with two homophones, prompt the participant with a question and ask which is the correct homophone (e.g., “Which belongs on your face?” *eye* vs. *I*). In both orthographic and

homophone choice tasks, the words, if sounded out, will be the same. Participants must rely on the orthographic features to determine the correct answer. Exception word reading tasks require participants to read exception words (e.g., *salmon*) out loud. Because these words are not regular, in that their pronunciations do not match their grapheme representation, participants cannot get the correct answer by phonological recoding.

Research on cumulative orthographic knowledge has found that it is a significant predictor of word identification (Holland et al., 2004), even after statistically controlling for age, IQ, and phonological abilities (Barker et al., 1992; Cunningham et al., 2001; Cunningham & Stanovich, 1990). Further, these results have been seen in studies including participants ranging from first through sixth grades (Barker et al., 1992; Cunningham et al., 2001; Cunningham & Stanovich, 1990; Holland et al., 2004) and whether predicting untimed or timed word identification (Barker et al., 1992). Research has also indicated that cumulative orthographic knowledge is a significant predictor of oral reading rate, silent reading rate, phonological recoding and later orthographic learning (Barker et al., 1992; Cunningham, 2006). Finally, cumulative orthographic knowledge also correlates with overall cognitive ability, receptive vocabulary, visual processing, RAN, phonological processing, and exposure to print (Bowers, Sunseth, & Golden, 1999; Conrad & Levy, 2007; Cunningham, 2006; Cunningham et al., 2001; Cunningham, Perry, Stanovich & Share, 2002; Cunningham & Stanovich, 1990; Georgiou, Parrila, Kirby, & Stephenson, 2008; Manis et al., 1999; Manis et al., 2000; Mesman & Kibby, 2011).

Current orthographic learning is best understood in the context of the self-teaching hypothesis. The self-teaching hypothesis (Jorm and Share, 1983; Share, 1995; 1999) predicts that orthographic learning is achieved through the process of phonological recoding. When

individuals phonologically recode new words, they learn the orthographic structures of those words as a byproduct. To test this hypothesis, self-teaching studies typically have participants read aloud nonwords (e.g., *yait*) and are then tested on their orthographic learning of those nonwords days later using multiple choice, spelling and reading reaction time tasks. The multiple choice tasks present participants with four spellings of the target and ask the participants to choose which one they learned days prior. These alternatives typically include the target (*yait*), a homophone (*yate*), a visually similar foil (*yoit*), and a letter transposition foil (*yiат*).

A classic study by Share (1999) provided evidence in support of the self-teaching hypothesis with second-grade participants who read aloud short texts with embedded nonword targets. Results of Share's study indicated that three days later, participants chose the nonword target more often than alternative foils, read the nonword targets more quickly than homophones and real words, and spelled the nonword targets more accurately than alternative homophones. In follow-up studies, Share created conditions that limited the opportunity for phonological recoding by either presenting the target very briefly (only 300 milliseconds) or by having participants vocalize concurrently by saying "dubba" repeatedly while the nonword target was presented. In both of these conditions, the opportunity to phonologically recode was minimized, and orthographic learning was significantly diminished.

Additional empirical tests of the self-teaching hypothesis have yielded supportive results for children ranging from first through fifth grades (Cunningham, 2006; Cunningham et al., 2002; Share, 2004) and in a variety of languages (Nation, Angell, & Castles, 2006; Share, 2004). These results have also upheld for silent reading (Bowery & Muller, 2005; de Jong & Share, 2007) and regardless of whether targets were presented individually or embedded in stories or sentences, thereby providing context (Nation et al., 2006; Share, 1999). Notably, Nation and

colleagues found that while learning increased with exposure, orthographic learning was still achieved after phonologically recoding a target only once, and Share (2004) found that orthographic learning of a set of targets was maintained up to one month later. Further support of the self-teaching hypothesis includes studies that have found that children who are better at phonological recoding show more orthographic learning (Cunningham, 2006; Cunningham et al., 2002; Oullette & Fraser, 2009). While understudied in comparison to phonological processing, orthographic processing clearly has a role in reading development.

### **Reading Skills in Down Syndrome**

Estimates of literacy rates and reading abilities for individuals with DS are not known, nor is the extent to which these individuals can master reading. Studies vary in their reports of the number of participants with DS who can and cannot read, and many studies may focus only on a subset of individuals who have some reading ability (Bochner & Pietrese, 1996; Buckley & Bird, 1993; Cardoso-Martins & Frith, 2001; Carr, 1988; Fidler et al., 2005). Abbeduto, Warren and Connors (2007) predict that these numbers vary from study to study based on cohort, recruiting strategies, and educational opportunities dependent on location.

The present study will address this issue by first surveying a large sample of parents of children with DS about their children's reading skills. This will be part one of the study and will describe the overall reading abilities of a sample of individuals with DS. Then, I will draw a sample of individuals with DS from the larger set to test their reading skills. This will be part two of the study. By using this two-part approach, the results of part two can be placed in a larger context (part one), which previous studies have not been able to do.

One issue involved in research on ID in general is matching. When studies match groups or participants on one variable, they will be mismatching on another. For example, within

reading research on DS it is typical to match a group with DS to a TD group on word identification ability (see Gombert, 2002; Roch & Jarrold, 2008; Verucci, Menghini, & Vicari, 2006), but to match on this variable requires that the groups be mismatched in chronological age (an older group of participants with DS is needed to perform equivalently on word identification), not to mention IQ. As Silverman (2007) notes, “it seems imprudent to assume that an 18 year-old person with DS should be qualitatively indistinct from a TD 8 year-old” (p. 233). This limitation is common knowledge within the field of intellectual and developmental disabilities research, yet no perfect solution has been found. Research on DS continues to match on single variables, thereby mismatching on others.

Research on reading skills in DS is sparse, especially in contrast to research on reading in the typical population. Using the Simple View of Reading (Gough & Tunmer, 1986) as an outline, below I report the research that has been done on various reading skills in DS. Generally, research on reading comprehension in DS is limited; however, due to known impairments in speech and language, more attention has been paid to the language comprehension domain in DS. Research in the word identification domain has primarily focused on word identification and phonological recoding. Almost no research has examined orthographic processing in DS.

Research on reading comprehension in DS is especially limited, but what is available suggests it is an area of significant difficulty (Fowler, Doherty, & Boynton, 1995; Moni & Jobling, 2001; Verucci, et al., 2006). Studies on reading comprehension have shown impairments in comprehension beyond what would be expected based on age and word identification ability (Byrne, Buckley, MacDonald, & Bird, 1995; Byrne, MacDonald, & Buckley, 2002; Cardoso-Martins, Peterson, Olson, & Pennington, 2009; Fowler et al., 1995;

Groen, Laws, Nation, & Bishop, 2006; Verucci et al., 2006). Research has also indicated that even with instruction, reading comprehension in DS progresses very slowly and lags behind that of TD children matched on word identification ability (Byrne et al., 2002).

Research with children with language impairments has indicated that language ability is related to current and future reading ability (Catts, Fey, Tomblin, & Zhang, 2002; Feagans & Appelbaum 1986; Snow, Tabors, Nicholson & Kurland, 1995). As noted earlier, language abilities of individuals with DS are poor for chronological age, mental age, and IQ. Research also indicates that individuals with DS typically perform worse on language measures in comparison to younger TD children matched on word identification ability (Byrne et al., 2002; Byrne et al., 1995; Hulme et al., 2012). Hulme and colleagues' recent study reported large effect sizes for the differences between a group with DS and a TD group matched on word identification ability for expressive ( $d = 1.14$ ) and receptive ( $d = 1.18$ ) vocabulary. Because of these severe deficits, language has been reported to be an even stronger predictor of reading ability in DS than in typical development (Boudreau, 2002).

Alternatively, research on word identification suggests that this is an area of strength for individuals with DS (Cupples & Iacono, 2000; Fowler et al., 1995) and is typically higher than what might be expected based on mental age (Cupples & Iacono, 2000; Fowler, Doherty, & Boynton, 1995; Kay-Raining Bird, Cleave, & McConnell, 2000), at least when those mental ages are 8 years or younger. Studies have also found that individuals with DS often outperform TD children matched on nonverbal ability and mixed ID samples matched on chronological age and IQ (Boudreau, 2002; Fidler et al., 2005). Further, some studies have indicated that word identification ability even surpasses overall cognitive functioning in individuals with DS (Byrne et al., 1995; Byrne et al., 2002).

Although not included in the Simple View (Gough & Tunmer, 1986), an obvious variable to consider in reading research on DS is IQ. The available research on the relationship between intelligence and reading is scarce. However, some research on TD samples has identified that general intelligence is a significant predictor of reading ability (Ellis & Large, 1988; Stanovich, Cunningham & Feeman, 1984), and some research on DS has also found relationships between intelligence and reading (Cardoso-Martins et al., 2009; Carr, 1995; Sloper, Cunningham, Turner, & Knussen, 1990). In their study, Cardoso-Martins and colleagues found significant differences in IQ between groups of strong and poor readers with DS, based on word identification ability. Other research with participants with ID has failed to find significant relationships between reading and full-scale IQ (Conners, 1990; Slate, 1995). However, when conducting research on any cognitive abilities in DS, including reading, it is good practice to include a measure of IQ.

**Phonological processing.** Research on phonological recoding in DS has identified interesting patterns, which appear to indicate poorer phonological recoding relative to better word identification abilities (Hulme, et al, 2012; Kay-Raining Bird et al., 2000; Næss, Melby-Lervag, Hulme, & Halaas Lyster, 2012a; 2012b). Previous research has found that individuals with DS struggle a great deal with phonological recoding (Cupples & Iacono, 2000; Kay Raining-Bird et al., 2000; Roch & Jarrold, 2008; Verucci et al., 2006). A meta-analysis by Næss and colleagues (2012a; see also Næss et al., 2012b) comparing phonological recoding in individuals with DS to TD peers matched on word identification ability found a moderate to large aggregated effect size (Hedges  $g = -.89$ ,  $p < .01$ ), indicating that individuals with DS struggle with phonological recoding, even relative to their word identification ability.

Two longitudinal studies by Hulme et al. (2012) and Kay-Raining Bird et al. (2000) have also found that phonological recoding does not keep pace with word identification skills in a DS

sample. In a two-year study, Hulme and colleagues examined reading, including measures of phonological recoding and word identification, in a sample of 45 individuals with DS ranging from Grades 1 to 12 and in a sample of TD participants ranging from Kindergarten to Grade 5. Groups were matched on word identification ability at Time 1. Each participant was measured three times on word identification (Times 1, 2 and 3) and two times on phonological recoding (Times 2 and 3). Results indicated that at both Times 2 and 3, the individuals with DS had significantly weaker phonological recoding skills than would be expected based on their word identification ability.

A second influential study by Boudreau (2002) showed that when a sample of participants with DS was superior to a TD group on word identification, there were no group differences on phonological recoding. Boudreau's study examined reading skills in a sample of 20 children and adolescents with DS compared to a sample of TD participants matched for nonverbal cognition. Results found no group differences on measures of early literacy variables (including tasks of letter name and letter knowledge). A significant group difference was found for word identification, with the group with DS performing better than the TD group. However, no group differences were found in phonological recoding.

Two important subskills of phonological recoding also appear to be problematic for individuals with DS. Both phonological awareness and phonological memory have been found to be impaired in individuals with DS (Boudreau, 2002; Cossu, Rossini, & Marshall, 1993; Fowler et al., 1995; Kay-Raining Bird et al., 2000; Lemons & Fuchs, 2010). Research examining phonological awareness in DS has indicated deficits when compared to TD controls matched on reading ability (Cardoso-Martins & Frith, 2001; Cardoso-Martins, Michalick, & Pollo, 2002; Cossu et al., 1993; Gombert, 2002; Roch & Jarrold, 2008; Snowling, Hulme, &

Mercer, 2002), even when controlling for cognitive ability (Snowling et al., 2002; Verucci et al., 2006) and when compared to controls matched on mental-age (Boudreau, 2002) or chronological age (van Bysterveldt, Gillon, & Moran, 2006). The deficit in phonological awareness has also been found across a huge age range in DS, from as young as 5.5 years upwards to 49 years (Boudreau, 2002; Cardoso-Martins & Frith, 2001).

One much-debated study by Cossu et al. (1993) found that a group of 10 children with DS performed worse than younger TD children matched on word identification ability (approximately 7 years) on four measures of phonological awareness. Cossu and colleagues concluded that if individuals with DS can read words despite very low performances on measures of phonological awareness, then phonological awareness must not be necessary to learn to read. However, Cossu and colleague's study has been criticized for both the methodology employed and their interpretation of results (see Byrne, 1993; Fletcher & Buckley, 2002). First, three of the four phonological awareness tasks required vocal production by the participant, which could have been influenced by poor speech abilities in the group with DS. Second, while some participants did perform at floor on a couple of the phonological awareness tasks, most of the participants did not. Further, for the one task not involving speech production, only one of the 10 participants with DS scored at floor. Critics of Cossu and colleagues assert that it was therefore premature to claim individuals with DS have no phonological awareness ability and that learning to read takes place in the absence of phonological awareness.

Indeed, several studies have contested Cossu and colleague's (1993) findings and found that phonological awareness is important to reading development in DS (Cardoso-Martins & Frith, 2001; Fower et al., 1995; Gombert, 2002; Snowling et al., 2002; Verucci et al., 2006). Further, in a study of 33 participants with DS described as "readers", Cardoso-Martins and Frith

(2001) found differences between production and recognition-based phonological awareness tasks. While still impaired when compared to reading matched TD participants (matched on both word identification and phonological recoding) on two measures of phonological awareness, the group with DS performed much better on a recognition measure than on a production measure.

Several researchers have noted and hypothesized that individuals with DS may learn to read differently than the general population. Buckley (1985) proposed that individuals with DS learn to read visually, skipping the letter-to-sound relationship between words. Buckley supported her claim with research showing that individuals with DS make semantic (i.e., *dog* for *cat*), rather than phonological (i.e., *car* for *cat*), errors when reading. Hulme et al. (2012) concluded that reading development in DS has both similarities and differences to the pattern observed in TD children. Hodapp and Fidler (1999) recommended a visual approach to teaching word identification skills to individuals with DS, and Abbeduto et al. (2007) proposed that individuals with DS may have stronger visual skills which contribute to their strengths in word identification. Since both Buckley and Cossu and colleagues' (1993) hypotheses were originally proposed, research has indicated that phonological processing is relevant to reading development in DS. However, phonological processes do not tell the full story of reading in DS, and research cannot ignore the role of orthographic processing, especially considering visual strengths for individuals with DS.

**Orthographic processing.** Despite its importance to reading, orthographic processing has not been researched as broadly as phonological recoding, even within the TD literature. The research on orthographic processing in DS is limited to cumulative orthographic knowledge using only exception word reading tasks. While these do tap orthographic knowledge, they are

production, as opposed to recognition measures. No studies have yet used orthographic awareness or orthographic choice tasks to examine orthographic knowledge in a DS sample.

*Production measures of orthographic knowledge.* The studies of exception word reading in DS have consistently found that individuals with DS show evidence of exception word reading ability (Fletcher & Buckley, 2002) and perform on par with younger, TD control groups matched on word identification accuracy (usually around a 7 year-old reading ability) and speed (Gombert, 2002; Roch & Jarrold, 2008; Verucci et al., 2006), despite lower mental ages (Cardoso-Martins et al., 2009; Cossu et al., 1993; Verucci et al., 2006). These results have been seen for a wide range of ages in DS, from 6 years upwards to young adults (Roch & Jarrold, 2008; Verucci et al., 2006) and across three different orthographies – English, Italian and French (Cardoso-Martins et al., 2009; Verucci et al., 2006; Gombert, 2002).

A study by Cardoso-Martins and colleagues (2009) found that individuals with DS and TD controls performed similarly on a measure of exception word reading. In their study, Cardoso-Martins and colleagues examined regular and exception word reading in 19 American, English-speaking adolescents with DS (mean age = 14.48; range = 10-19) in comparison to a group of younger TD children (mean age = 8.43; range 8-9) and to a second group of participants with dyslexia (mean age = 10.64; range 8-15), all matched on word identification ability. Despite an average mental age of 4.9 years for the group with DS, all groups, including the group with DS, had word identification abilities at a third to fourth grade reading level. There were no significant differences between groups on exception word reading. Results also indicated that all groups performed better on regular word reading than exception word reading. Interestingly, though, while there were no group differences on the regular word reading task, the TD and dyslexia groups both had larger discrepancies between the two tasks than did the group with DS.

Another study by Roch and Jarrold (2008) found that individuals with DS out-performed TD controls on a measure of exception word reading. Further, the magnitude of this effect was fairly large (Cohen's  $d = .78$ ). Roch and Jarrold conducted a similar study to that of Cardoso-Martins and colleagues (2009), but reported only including participants with stronger reading skills (those whose skills surpassed letter identification). Twelve participants with DS (mean age = 18.92, range 10.42-26.58) were selected to participate. These participants were matched to younger TD participants (mean age = 6.83, range 6.25-7.25) based on word identification ability. Roch and Jarrold had participants read aloud 20 one- and two-syllable exception words. Results of their study indicated that the group with DS performed significantly better on the exception word reading task in terms of accuracy, but there were no significant differences between groups for speed. In contrast to Cardoso-Martins et al. (2009), Roch and Jarrold reported a trend in which the group with DS performed slightly better on exception word reading vs. a regular word reading task.

While Roch and Jarrold (2008) found that their group with DS performed better on an exception word reading task than did the younger TD participants, the majority of research on exception word reading in DS has failed to find significant effects between these two groups. Instead, the literature is more consistent with Cardoso-Martins and colleagues (2009) who found no significant differences between groups of students with DS and TD students matched on word identification level (Cossu et al., 1993; Gombert, 2002; Verucci et al., 2006). Participants in Roch and Jarrold's study had higher mental ages (8 years vs. 4.9 years in Cardoso-Martins et al.'s study) and were compared to slightly younger TD participants (mean = 6.83 years vs. 8.43 years in Cardoso-Martins et al.'s study). Although not reported, it is also possible, based on

mental ages, that Roch and Jarrold's participants with DS had slightly higher IQs than those in the other studies.

Taken together, results of exception word reading studies suggest that individuals with DS are reading exception words on par with their word identification ability, but better than what would be expected based on mental age. Further, older individuals with DS, possibly those who have more experience with print, may even have an advantage in exception word reading (Roch & Jarrold, 2008). These results indicate that orthographic knowledge may be an area of strength, relative to phonological recoding, for individuals with DS. Results of these studies should be interpreted with caution, though, as they have had samples as small as 10.

***Recognition measures of orthographic knowledge.*** While no studies have examined cumulative orthographic knowledge in DS samples using orthographic awareness and orthographic choice tasks, a few studies have done so in ID samples. Using an orthographic awareness task in which participants were presented with two nonwords (e.g., *pokerson* vs. *bhdtunkqk*) and asked to select which one looks more like a real word, Allington (1981) found that 30 children with mild ID (mean IQ = 76) showed evidence of orthographic knowledge. Results from Allington's study also revealed that performance on the orthographic awareness task was related to performance on word identification and reading comprehension tasks.

Two additional studies measuring orthographic processing in ID also indicate that this skill may be relatively good when compared to phonological recoding. Channell, Loveall, and Connors (2013) found that a group of participants with ID ranging from sixth to 12<sup>th</sup> grade performed as well as TD, mental-age matched peers on two different orthographic knowledge tasks: orthographic awareness and homophone choice. In a separate study, Loveall and Connors (2013) found that a group of adolescent and adult participants with ID were able to learn

orthographic structures of nonwords as well as TD participants of the same verbal mental age by sounding out each nonword. Based on these studies, one could expect participants with DS to also perform better on measures of orthographic knowledge than phonological recoding.

The available literature on orthographic processing in DS is limited to cumulative orthographic knowledge and suggests relatively strong performances on exception word reading tasks. However, as noted before, exception word reading tasks are production, as opposed to recognition, tasks. Individuals with DS may struggle with production tasks due to poor language abilities (Abbeduto et al., 2003) and poor working memory (Jarrod & Baddeley, 2001). With strengths in visual processing (Buckley, 1985; Connors et al., 2011b), individuals with DS may show stronger evidence of orthographic knowledge using recognition tasks, such as orthographic awareness and orthographic choice. It is possible that their performance on these measures could surpass what would be expected based on their IQ, reading ability and mental age, as they are recognition and not production tasks.

Taken together, the literature on reading in DS suggests that when matched on word identification ability to younger TD participants, individuals with DS will show lower mental ages (e.g., Cardoso-Martins et al., 2009), lower IQs (e.g., Gombert, 2002) higher chronological ages (e.g., Cardoso-Martins et al., 2009) and lower phonological processing skills (e.g., Kay-Raining Bird et al., 2000). Performance on orthographic processing tasks could be on par (e.g., Gombert, 2002) or even better (e.g., Roch & Jarrod, 2008) in individuals with DS when compared to TD participants matched on word identification ability.

### **Predictors of Reading Development**

Environmental factors have also been acknowledged as significant precursors to reading development in TD children, and it is possible that these factors also relate to reading

development in DS. The home literacy environment is defined as the frequency and nature of literacy-related activities in the home. It is measured by parent questionnaires examining the onset, frequency and quality of parent-child shared book reading, the number of books in the home, the frequency of trips to the library, and the frequency of reading by caregivers. Research with TD children has found that the home literacy environment predicts later language development, emergent literacy, and later reading achievement (Bus, van Ijzendoorn & Pellegrini, 1995; Lyytinen, Laakso & Poikkeus, 1998; Scarborough & Dobrich, 1994; Senechal, 1997). Richer home literacy environments, for example characterized by more parent-child shared book reading or more books in the home, are believed to contribute to language and reading achievement because they expose children to a wider variety of sentence forms than would be experienced through spoken language alone, can help make children more aware of letter-sound correspondences, and expose children to story structures and literacy conventions (Bus et al., 1995; DeBaryshe, 1993; Tannen, 1982).

A few studies have examined the home-literacy environment in DS (Al Otaiba, Lewis, Whalon, Dyrland, & McKenzie, 2009; Fitzgerald, Roberts, Pierce, & Schuele, 1995; Ricci, 2011), but only one included a TD control group (Ricci, 2011). Results from Ricci's study suggest that grade-school age children with DS (8-13 years) and TD pre-school age children (3-5 years) were exposed to more rich home literacy environments than pre-school age children with DS (3-6 years). Ricci also reported no significant differences in vocabulary between the grade-school children with DS and the TD group. With such limited research, it is difficult to know how the home literacy environment relates to reading in DS. It is possible that the home literacy environment influences reading development differently in DS than in the typical population. No studies have yet examined the relation of the home literacy environment to word

identification, phonological recoding or orthographic knowledge in a DS sample. The present study seeks to examine the home literacy environment and its relation these reading skills in DS.

Rich home literacy environments have also been linked to increases in TD children's interest in reading (Lyytinen, Laakso & Poikkeus, 1998). Within TD children, motivation to read has been found to predict both the amount and breadth of reading (Wigfield & Guthrie, 1997). If a child is interested and motivated to read, then he/she may ask parents to read with them more when young and/or read more themselves as they develop their own reading skills. They may also be exposed to a greater breadth of reading material. This exposure to, and experience of, reading when young could then contribute to later reading achievement. In her study, Ricci (2011) reported that the grade-school children with DS and the TD group (who had richer home literacy environments) indicated more interest in reading than pre-school age children with DS. No other studies have examined motivation to read within a DS sample, and no studies have linked motivation to read with word identification, phonological recoding or orthographic knowledge in DS. The present study seeks to measure the home literacy environment and interest in reading to explore the relations of these factors with reading development within a DS sample.

## **Summary**

The DS cognitive profile is associated with poor verbal abilities, but relatively stronger visuospatial abilities (Conners et al., 2011b). Previous research on reading in Down syndrome has illustrated word identification abilities that are higher than what might be expected based on mental age (i.e. Cupples & Iacono, 2000). This is somewhat surprising considering research has also indicated that one of the main subskills of word identification, phonological recoding, is impaired in DS (i.e. Hulme et al., 2012). It is possible then that individuals with DS, capitalizing

on their visuospatial skills, have developed stronger orthographic processing skills to use when reading words. However, there is little to no research examining orthographic processing in DS.

### **Proposed Study**

The main purpose of the present study was to examine cumulative orthographic knowledge in individuals with DS. In many reading studies individuals with DS have performed better on measures of word identification than on measures of phonological recoding. If phonological recoding is impaired, it is possible that orthographic processing, the visual subskill of word identification, is relatively strong. Abbeduto et al. (2007) noted that the discrepancy between word identification and phonological recoding might be explained by stronger visual skills through the process of “matching orthographic patterns”. However, research has yet to really examine orthographic processing in a DS sample. In the present study, orthographic knowledge of individuals with DS was examined and compared to those of TD children matched on word identification ability. A second purpose of the present study was to determine literacy rates of individuals with DS. Previous studies have varied in their reports of reading ability in DS, and many of these studies failed to report details of their samples, possibly only focusing on individuals with DS who have some reading ability. The current study used parent reports to examine literacy rates in a DS registry sample. Also from the parent reports, individuals with DS reported to have reading abilities were recruited in order to examine their orthographic knowledge.

**Part 1: Parent survey.** The parent survey study attempted to report literacy rates and varying reading abilities in a DS registry sample. All parents registered who have a child with DS were contacted for participation. Parents were asked to report on their child’s reading ability, their children’s motivation to read and the home literacy environment. Reading questions were

designed to quantify the percentage of individuals with DS within different age groups, who 1) know the letters of the alphabet, 2) can sound out unfamiliar words, 3) can read individual words, 4) can read short stories/magazines/books and 5) would be described as a reader by their parent. Further, information from the parent survey was used to explore the effects of motivation and the home literacy environment on reading development in DS.

**Part 2: Comparative child study.** The comparative study was the primary focus of the present study. The goal of this study was to examine reading abilities, especially cumulative orthographic knowledge, in individuals with DS in comparison to TD children matched on word identification ability. Participants with DS are compared to TD children on three measures of orthographic knowledge (two recognition tasks, one production task), two measures of word identification and one measure of phonological recoding. Participants also completed an IQ test and a measure of receptive vocabulary to allow for an examination of how these skills relate to reading abilities within and across each sample.

## **Hypotheses**

### **Primary Hypotheses**

1. Overall, the group with DS will perform better than the TD group on cumulative orthographic knowledge.
2. The group with DS will perform worse than the TD group on phonological recoding.

The first hypothesis, that the group with DS will perform better than the TD group on measures of orthographic knowledge, is built off of previous research suggesting that individuals with DS have relatively strong orthographic skills. When using exception word tasks, which require a verbal production by participants, several studies have found that individuals with DS perform on par with younger TD participants matched on word identification ability (Cardoso-

Martins et al., 2009; Cossu et al., 1993; Gombert, 2002; Roch & Jarrold, 2008; Verucci et al., 2006). One study by Roch and Jarrold (2008) found that a group of individuals with DS outperformed a TD group matched on word identification ability. The present study will include Exception Word Reading (a production task), as well as two additional measures of orthographic knowledge, both recognition tasks. Based on previous research indicating poor language and poor working memory (Abbeduto et al., 2003; Cain et al., 2004), but stronger visual skills (Buckley, 1985; Connors et al., 2011b), I predict that individuals with DS will perform better on orthographic knowledge tasks overall when recognition measures are included in the protocol. The second hypothesis, that the group with DS will perform worse than the TD group on phonological recoding, will replicate previous research (see Næss et al., 2012a; 2012b) indicating that individuals with DS are poor in phonological recoding compared TD peers matched on word identification ability.

### **Exploratory Hypotheses**

In addition to the primary hypotheses above, I hypothesize:

3. An interaction between group (DS vs. TD) and measure of orthographic knowledge (recognition vs. production). The two groups will perform similarly on a production measure of orthographic knowledge, but the group with DS will perform significantly better on recognition measures.
4. An interaction between group (DS vs. TD) and subskill of word identification (phonological recoding vs. orthographic knowledge). Exception Word Reading will be used as the measure of orthographic knowledge, as both phonological recoding and Exception Word Reading are production measures. I hypothesize that the two groups

will perform similarly on Exception Word Reading/ orthographic knowledge, but the TD group will perform significantly better on phonological recoding.

5. Receptive vocabulary will correlate with participants' reading abilities, including orthographic knowledge, in both groups.
6. Using data from the parent survey along with data from participants with DS in the comparative child study, relationships between parents' reports of child motivation to read and the home literacy environment will be examined to see if any significant relationships exist between these variables and the participants' performances on various reading measures.

## METHODOLOGY

### Participants

**Parent survey study.** Out of approximately 140 families from the University of Alabama Intellectual Disabilities Participant Registry (UAIDPR) invited to participate in the study, 56 were interested in participating, completed the study and are included in data analyses. The UAIDPR is a registry of families in Alabama, Mississippi, Georgia, and Florida who have a child with ID, whether with or without DS, and who are interested in participating in research projects focused on ID. There are approximately 140 families in the registry who have a child with DS. All parents of a child with DS in the registry were contacted. Those who agreed to participate were offered the option of 1) receiving the questionnaire via mail with pre-paid return postage or 2) completing the questionnaire via phone with an experimenter. See Table 1 for demographics of children whose families participated in the study.

Table 1

*Children's Demographics*

	<i>n</i>	%
Gender		
Male	26	46.4%
Female	30	53.6%
Age		
2-5 years	8	14.3%
6-9 years	10	17.9%
10-12 years	6	10.7%
13-15 years	10	17.9%
16-19 years	6	10.7%
20-29 years	12	21.4%
30-45 years	4	7.1%

*Note: n = 56.*

**Comparative child study.** A total of 24 participants with DS and 26 TD participants participated in this study. In the group with DS, two participants did not meet the eligibility criteria of reading at least one word on each Word Identification subtest. Two other participants were excluded from data analyses due to behavioral concerns during testing. In the TD group, 20 out of the 26 total participants tested were selected for data analyses to make the best word identification match. Thus, the final sample included 20 participants with DS and 20 TD participants.

All of the participants with DS came from the UAIDPR. Their primary caregiver had 1) participated in the parent survey study, 2) reported that their child with DS could read single words and 3) reported that they at least somewhat considered their child a reader. Additional participation criteria for the group with DS included a raw score of at least one on each of two Word Identification subtests of the Woodcock Reading Mastery Test – Third Edition (WRMT-III). Using raw scores from each subtest, age-equivalence scores were calculated. The two age-equivalence scores were then averaged to obtain an aggregated word identification age-equivalent. TD participants were then recruited from local city schools to match the group with DS on word identification ability. Additional inclusion criteria for the TD participants included: no diagnosis of ADHD, not eligible for special education services, no speech or language impairments, and English as a first language. These eligibility criteria were checked through parent report. A final inclusion criterion for the TD participants was an IQ within a range of 80-130, as measured by the Kaufman Brief Intelligence Test – 2 (KBIT-2).

The participants with DS varied in age from 11 to 21 years (grades 4 to post-high school). Although the study is referred to as a “child comparative study” it is important to note that several of the participants with DS were adolescents and young adults. The term “child” is used

to denote that these participants were children of the parents who participated in the parent survey study. This wide age range has been used in several studies examining literacy skills in DS (see Boudreau, 2002; Cardos-Martins et al., 2002; Fidler et al., 2005; Laws & Gunn, 2002) because of difficulty recruiting participants, wide variability in mental ages and because it represents the school age years when reading skills are taught. Of the 20 participants with DS included in data analyses, the average word identification age equivalence ranged from 6.50 to 9.96 years ( $M = 7.64$  years,  $SD = 1.01$ ). To match the group with DS on word identification ability, TD participants ranged in age from 5-9 years (Kindergarten to 3<sup>rd</sup> grade). Of the 20 TD participants included in data analyses, the average word identification age equivalence ranged from 6.33 to 10.83 years ( $M = 7.60$ ,  $SD = 1.18$ ).

The group with DS was 60% female, 40% male, 85% Caucasian, 10% Hispanic and 5% other. The TD group was 40% female, 60% male, 30% Caucasian, 45% African-American, 15% Hispanic and 10% other. Because the groups differed somewhat in gender, independent samples *t*-tests were used to compare male and female performance within each group (DS and ID) on each reading measure (word identification, phonological recoding and the three orthographic knowledge tasks). Results of the *t*-tests did not indicate significant differences between male and female performance in either group. Further, because the TD group was more racially diverse than was the group with DS, independent samples *t*-tests were also used to compare Caucasian and minority students on each of the reading measures. The only significant difference between the groups was on Orthographic Choice,  $t(18) = 2.28$ ,  $p = .04$ , with minority students ( $M = 58.07$ ,  $SD = 9.97$ ) performing significantly better than Caucasian students ( $M = 47.00$ ,  $SD = 9.94$ ). Also, within the TD group, all but one participant had word identification age-equivalence

scores consistent or slightly above their chronological age. Only one participant scored less than a year below his/her chronological age on word identification.

Data from the parent survey study also revealed that for participants with DS, 87.5% of those in the child study were reported to have some vision impairment, and 90% of these participants wore glasses or contact lenses. Eleven participants with DS were also reported to have some hearing loss (45.8%), and 27% of these participants used a hearing aid. See Table 2 for sample descriptives.

Table 2

*Means and Standard Deviations of Sample Descriptives*

	DS <i>n</i> = 20		TD <i>n</i> = 20	
	Mean	SD	Mean	SD
Chronological Age	16.16	3.33	7.33	.97
IQ	47.75	8.83	103.30	12.31
Word ID (A) Raw Score	18.50	6.01	17.95	7.17
Word ID (A) Age Equivalence	7.68	1.13	7.72	1.44
Word ID (B) Raw Score	17.45	5.63	16.65	5.58
Word ID (B) Age Equivalence	7.60	.98	7.48	.97
Word ID Average Age Equivalence	7.64	1.01	7.60	1.18

**Design**

**Parent survey study.** Descriptive statistics were used to estimate the literacy rates in a DS registry sample. Based on parent report, percentages of several reading abilities were calculated, including letter identification, phonological recoding, and word identification. Descriptive statistics were also used to describe the home literacy environment and the child’s motivation to read.

**Comparative child study.** To investigate cumulative orthographic knowledge in a sample of participants with DS, this study utilized a one-way multivariate analysis of variance.

The between groups variable was group (DS vs. TD), and the combined dependent variables included performance on Orthographic Awareness, Orthographic Choice and Exception Word Reading. Follow-up univariate analyses were also used to compare groups on each individual orthographic measure. A Mann-Whitney U test was used to compare groups on phonological recoding.

### **Measures: Parent Survey Study**

**Parent Reading Questionnaire** (15 min.; see Appendix B) is a parent questionnaire designed by our lab, which includes questions about the parent's perception of their child's reading ability, the home literacy environment and the child's motivation to read. This questionnaire was adapted from Al Otaiba et al. (2008), Ricci (2011), and van der Schuit, Peeters, Segers, Balkom, and Verhoeven (2009). In addition, parents were asked about their child's hearing, vision and language. Percentages of individual items were calculated as were sum scores for each of three categories: reading ability, home literacy environment and reading motivation. Both percentages of individual items and sum scores were used in the present study.

The questionnaire had good internal consistency. For the reading abilities subscale, consisting of 11 items, the Chronbach alpha coefficient was .95. For the home literacy environment subscale, consisting of 13 items, the Chronbach alpha coefficient was .74, and for the reading motivation subscale, consisting of 4 items, the inter-item correlation ranged from .29-.79.

### **Measures: Comparative Child Study**

**Woodcock Reading Mastery Tests- Third Edition** (WRMT-III; Woodcock, 2011; 15 min.) is a test of specific reading skills and overall reading. It provides raw scores, age-equivalences, grade-equivalences, standard scores, and growth score values for each individual

subtest and composite scores for reading readiness, basic reading skills, reading comprehension and total reading ability. Raw scores, age-equivalences and aggregated age-equivalences were used in the present study to match participants from each group.

The WRMT-III comes in two forms, A and B. Both forms were used in the present study. From Form A, two subtests were used — Word Identification, which measures reading of real words, and Word Attack, which measures nonword reading (phonological recoding). The Word Identification subtest from Form B was used in addition to the Word Identification subtest from Form A to match participants on word identification ability. The Word Identification subtest consists of 46 isolated words of increasing complexity. Items are presented on easel pages, and participants are asked to read the words aloud. The Word Attack subtest consists of 26 nonwords (i.e. *dee, ap*) of increasing complexity. Items are presented on easel pages, and participants are asked to read the nonwords aloud. Because these are not real words, or in some cases are words of very low frequency in the English language, participants should not have been exposed to them before and therefore must phonologically recode the items in order to read them aloud. Because participants with DS have poor speech, including poor articulation and/or difficulty with specific phonemes, the WRMT-III was audio-recorded. This allowed examiners to re-listen to items and consult with a second investigator to score difficult items.

The WRMT-III is a widely used norm-referenced battery that is appropriate for ages 4.5 years to 75+ years. For the Word Identification subtests, split-half reliability ranges from .85 to .98 for all ages and from .97-.98 for the youngest participants reported, who are 6 years old. For the Word Attack subtest, Form A, split-half reliabilities are also strong, ranging from .75-.95 for all ages and .94 for the youngest participants reported. For Pre-Kindergarten to Grade 6, the WRMT-III also correlates with the Woodcock-Johnson Tests of Achievement .81 for Word

Identification and .82 for Word Attack (Woodcock, 2011). The WRTM-III reports that children in Grade 1 receive an average raw score of approximately 16 on each Word Identification subtest and an average raw score of approximately 9 on the Word Attack subtest, Form A. Forms A and B correlated at .89 in this study's sample.

**Kaufman Brief Intelligence Test – 2** (KBIT-2; Kaufman & Kaufman, 2004; 20 min.) is a brief measure of both verbal and nonverbal intelligence. Two subtests, Verbal Knowledge and Riddles, together provide a verbal standard score, and a third subtest, Matrices, provides a non-verbal standard score. The KBIT-2 also yields raw scores, age-equivalences and an IQ composite. The Verbal Knowledge subtest consists of 60 items and measures receptive vocabulary and general information about the world. For each item, six pictures are presented on a page. Participants are asked to pick which of the six pictures shows the meaning of the word or answers the question spoken by the examiner. The Riddles subtest consists of 48 items and measures verbal comprehension, reasoning and vocabulary knowledge. For the first eight items participants are asked to pick which of eight pictures answer a riddle spoken by the examiner. For the remaining 40 items participants are asked to verbally respond with a single word that answers the riddle. The Matrices subtest consists of 46 items and measures nonverbal reasoning and understanding of relationships among stimuli, both meaningful and abstract. For the easiest items, participants are presented a stimulus picture and asked to select which of five alternative pictures goes best with the stimulus. For the next set of items, participants are asked to select which of six pictures best completes a 2x2 visual analogy. The most difficult set of items ask participants to select which alternative picture best completes a 2x2 or 3x3 matrix. The present study used IQ, verbal age equivalence scores as a measure of verbal mental age and nonverbal age equivalence scores as a measure of visual analysis skill.

The KBIT-2 is a standardized test that is appropriate for functioning levels equivalent from 4 years to 90+ years. Internal-consistency reliabilities for the full test range from .89 to .96 for all ages and is .89 for the youngest participants at 4 years. Test-retest reliabilities for the full test range from .88 to .92 for all ages and is .88 for the youngest participants reported, ages 4 to 12 years. The KBIT-2 also correlates with the Wechsler Intelligence Scale for Children – Fourth Edition (WISC-IV) at .77 for the full test, .79 for the verbal sections, and .56 for the nonverbal section (Kaufman & Kaufman, 2004).

**Child Reading Motivation Questionnaire** (2 min.; see Appendix C) this is a two-to-four item questionnaire developed by our lab that asks children how interested they are in reading. An experimenter read the questions to the participants and recorded their responses. Points were awarded for answers indicating interest and motivation in reading. Total scores could range from 0-6, with higher scores indicating more interest in and motivation to read. Chronbach's alpha for the questionnaire was .73 for the full sample, .67 for the sample of DS, and .85 for the TD sample.

**Orthographic Choice**, (Olson, Forsberg, Wise, & Rack, 1994), **Orthographic Awareness**, (Siegel, Share, & Geva, 1995; 15 minutes for both tasks) was used to measure orthographic knowledge via recognition. These two tasks are both computerized, were modified from Olson and colleagues (2004) and Siegel and colleagues (1995) and have previously been used in our lab with individuals with ID. For each orthographic task, accuracy was the primary measure. Raw scores out of 80 for Orthographic Choice and 67 for Orthographic Awareness were recorded by the computer and used in the present study.

The Orthographic Choice task measures knowledge of specific orthographic sequences in words by presenting word-nonword pairs such as snow-snoe one at a time on a computer screen.

Participants are asked to indicate the real word by pressing a corresponding key on the same side as their choice. Orthographic Awareness (modeled after Siegel et al., 1995) measures knowledge of general patterns of letters in words by presenting nonword pairs such as *filv-filk*. Nonword pairs (from Massaro, Taylor, Venezsky, Jastrzemski, & Lucas, 1980; Siegel et al., 1995; Treiman, 1993) are either linguistically legal or illegal, have regular or irregular spellings, and have high or low positional frequency of letters. Participants are asked to indicate which looks more like a real word by pressing a corresponding key on the same side as their choice. For the Orthographic Choice task, split-half reliability from a previous study was .72, and for the Orthographic Awareness task, split-half reliability was .85 (Connors et al., 2011a).

**Exception Word Reading** (10 min.; see Appendix A) was used to measure orthographic processing via production. This task consists of 25 exception words (i.e. *great, soul, island*) of varying frequency (with frequency ratings from 23 to 4,393 based on Kuchera & Francis, 1976) in the English language. All of the words violate at least one spelling-to-sound rule. Twenty exception words were taken from Roch and Jarrold (2008), and five additional, mostly higher-frequency exception words were added to the task. Items were presented individually on a page, and participants were asked to read the items out loud. Participants completed all 25 items, and raw score correct was used for the present study. Chronbach's alpha was .92 for the full sample, .91 for the group with DS and .94 for the TD group.

**Peabody Picture Vocabulary Test – 4<sup>th</sup> edition**, (PPVT; Dunn & Dunn, 2007; 15 min.), a measure of language ability, was used to measure receptive vocabulary. For each item, four pictures are presented on a page. Participants are asked to point to the picture that corresponds with a word spoken by the examiner. The test covers 20 content categories and includes nouns, verbs, and adjectives. Growth score values were used in the present study. Growth score values

are raw scores weighted for item difficulty. Higher scores indicate greater receptive vocabulary ability.

The PPVT is a standardized norm-referenced test that is appropriate for functioning levels equivalent from 2.5 years to 90+ years. Split half reliability ranges from .89 to .97 for all ages and from .95-.96 for the youngest participants reported (2:6-2:11). Test-retest reliabilities are also strong for the PPVT, ranging from .92-.93 for all ages and .93 for the youngest participants reported (ages 2-4 years). The PPVT also correlates with the Clinical Evaluation of Language Fundamentals, 4<sup>th</sup> edition (CELF-4) from .67 to .73 for ages 5-12 years, with the Expressive Vocabulary Test (EVT-2) from .80-.84 for ages 2-81+ years, and with the Comprehensive Assessment of Spoken Language (CASL) from .41 to .79 for ages 3-12 years (Dunn & Dunn, 2007).

## **Procedure**

**Parent survey study.** All parents of children with DS registered with the UAIDPR were contacted by the Registry Coordinator to see if they were interested in participating in the parent survey study. They were also told that some children from the survey study would be asked to participate in a follow-up child study. If parents indicated that they were interested in participating their information was given to the investigators. The primary investigator then contacted the parents via phone or e-mail to invite them into the study. Parents who wished to participate were given the option of completing the surveys via mail or over the phone with an investigator. Those who completed the survey via mail were sent the surveys along with instructions and a return envelope. Those who completed the study over the phone were read the instructions. The investigator then recorded verbal answers.

**Comparative child study.** Participants with DS were selected from the parent survey study based on parent reports of their reading ability. Only participants whose parents had indicated that they could read words and would be described as a reader were invited to participate in the child study. TD participants were recruited from local city schools. Participants were tested individually by a trained examiner in a quiet room. The full purpose of the study was disclosed to participants before testing. After obtaining both parental consent and child assent (child consent was also received for adult children with DS), the examiner administered the Word Identification subtests to see if participants were eligible for the study. If the participant was eligible the experimenter continued to administer the remaining tasks: Word Attack, KBIT-2, Motivation Questionnaire, Orthographic Tasks (in a balanced latin square design), and PPVT.

Testing occurred in one session, lasting from one to two hours, depending on the participant. At the end of the session, participants were debriefed, thanked and given the opportunity to ask any questions about the study. They were also given a small prize for participating.

## RESULTS

### Parent Survey Analyses

Fifty-six families participated in the parent survey study. These families all have a child with DS, ranging from 2 to 45 years of age. Most respondents were mothers (54 out of 56); one father and one grandmother completed the survey. All respondents were the primary caregiver of the child with DS. Most participants fully completed the questionnaires. However, some participants left select questions blank. To maximize *n*'s in every analysis, participants were not excluded from all analyses for leaving questions blank.

**Reading ability.** Descriptive statistics based on the parent survey were used to describe the literacy rates of a DS registry sample. Based on parent report, percentages of several reading abilities were calculated, including letter identification, phonological recoding, and word identification. Any participants younger than 6 years were not included when calculating percentages of reading abilities.

Especially relevant to the child study, a majority of parents (95.9%) reported that their child could read single words at least somewhat. A smaller percentage, but still majority, of parents (70.8%) reported that their child could sound out new/unfamiliar words at least somewhat. Twenty-seven percent of parents reported that they would not describe their child as a reader, 41.7% that their child is somewhat of a reader and 31.3% reported that they consider their child a reader. See Table 3 for a full summary of the results.

Table 3

*Parent Report of Child's Reading Abilities*

	No, not at all	Somewhat	Yes, very well
Recite the alphabet <i>n</i> = 46	4.3%	39.1%	56.5%
Identify single, written letters <i>n</i> = 48	2.1%	14.6%	83.3%
Write out letters <i>n</i> = 48	8.3%	25.0%	66.7%
Know sounds of letters <i>n</i> = 48	4.2%	33.3%	62.5%
Read single words <i>n</i> = 48	4.2%	29.2%	66.7%
Sound out new/unfamiliar words <i>n</i> = 48	29.2%	45.8%	25.0%
Read basic picture books or simple stories <i>n</i> = 48	14.6%	16.7%	68.8%
Read books that contain new words or concepts <i>n</i> = 48	25.0%	35.4%	39.6%
Read magazines <i>n</i> = 48	52.1%	27.1%	20.8%
Read news stories <i>n</i> = 48	66.7%	18.8%	14.6%
Would you describe your child as a reader? <i>n</i> = 48	27.1%	41.7%	31.3%

**Home literacy environment.** Descriptive statistics were also used to describe the home literacy environment for individuals with DS. These results include all participants, regardless of child's age. Almost all parents reported that there were some books in the home when their child was an infant, a toddler and currently (see Table 4). When the child was young most parents report reading to their child at least a few times a week (87.2% for infants, 96.4% for toddlers). Only 3.6% of parents reported that they never or almost never read to their child when their child was an infant or toddler. In contrast, 30.4% of parents reported never or almost never reading to their child currently. However, 60.6% still reported reading to their child at least a few times a

week (see Table 5). Further, most parents (76.4%) began reading to/with their child when their child was between birth and 6 months of age, and almost all parents (96.4%) began reading to their child before the child turned 1 year (see Table 5).

Table 4

*Number of Children's Books in the Home by Child's Age (reported in percentages)*

	None	0-50 books	50-100 books	100-200 books	Greater than 200 books
Infant <i>n</i> = 56	3.6%	37.5%	25%	17.9%	16.1%
Toddler <i>n</i> = 56	0%	26.8%	30.4%	26.8%	16.1%
Currently <i>n</i> = 56	5.4%	21.4%	21.4%	21.4%	30.4%

Table 5

*Time Parent Reads to/with Child by Child's Age (reported in percentages)*

	Never/ almost never	Weekly	A few times a week	Daily	A few times a day
Infant <i>n</i> = 55	3.6%	9.1%	32.7%	32.7%	21.8%
Toddler <i>n</i> = 55	0%	3.6%	23.6%	45.5%	27.3%
Currently <i>n</i> = 56	30.4%	8.9%	19.6%	30.4%	10.7%

*Age Parent Began Reading to Child (reported in percentages)*

	Not yet	Birth – 6 months	7-11 months	1-2 years	2-3 years	3-4 years	5-6 years	7 + years
<i>n</i> = 55	0%	76.4%	20.0%	1.8%	0%	0%	0%	1.8%

**Child's motivation to read.** Descriptive statistics were also used to describe child's reading motivation, as reported by the parent. These results include all participants, regardless of

child's age. A majority of parents (91%) reported that their child amuses him/herself with books, magazines, or comic books at least weekly. A majority of parents also reported that their child is interested in reading books with them (76.7%). See Table 6 for a full report of the results.

Table 6

*Child's Motivation to Read (reported in percentages)*

	Never/ almost never	Weekly	A few times a week	Daily	A few times a day
In a typical week, how often does your child amuse him/herself with books, magazines, or comic books?	8.9%	10.7%	23.2%	37.5%	19.6%
In a typical week, how often does your child ask you to read to him/her?	44.6%	3.6%	17.9%	25.0%	8.9%
		No interest	A little interest	A lot of interest	
How interested is your child in reading books with you?		23.2%	32.1%	44.6%	
How interested is your child in other reading activities?		8.9%	48.2%	42.9%	

*Note: n = 56.*

**Correlations.** For each of three categories from the parent survey (reading abilities, home literacy environment and child's motivation to read) sums were calculated to use as overall scores, with higher scores indicating stronger reading abilities, richer home literacy environments and higher motivation to read. These overall scores were used to examine the relationships between the child's age, reading ability, home literacy environment and reading motivation. These relationships were examined using Pearson product-moment correlation coefficients. If participants were missing data, they were only excluded for the particular

correlation in which data was missing. Results revealed a strong, positive relationship between age and reading ability,  $r = .55, p < .001$ , with increased age associated with increased reading ability. There was a medium, negative correlation between age and reading motivation,  $r = -.49, p < .001$ , with increased age associated with lower levels of reading motivation. Further, there was a medium, positive correlation between the home literacy environment and reading motivation,  $r = .42, p = .001$ , with richer home literacy environments associated with increased reading motivation. The relationship between age and the home literacy environment was non-significant,  $r = -.14, p = .32$ , as were the relationships between reading ability and reading motivation,  $r = -.16, p = .26$ , and reading ability and the home literacy environment,  $r = .04, p = .78$ . See Table 7 for correlations.

Partial correlations controlling for age were also used to explore the relationships among reading ability, the home literacy environment and reading motivation. Results revealed a positive, medium correlation between the home literacy environment and reading motivation,  $r = .41, p = .002$ , again with richer home literacy environments associated with increased reading motivation. The relationships between reading ability and the home literacy environment,  $r = .14, p = .32$  and reading ability and reading motivation,  $r = .16, p = .26$  were nonsignificant. See Table 7 for correlations.

Table 7

*Correlations between Age, Reading Ability, Home Literacy Environment and Reading Motivation (corrected for age below the diagonal)*

	Age	Reading Ability	Home Literacy Environment	Reading Motivation
Age	--	.55**	-.14	-.49**
Reading Ability	--	--	.04	-.16
Home Literacy Environment	--	.14	--	.42**
Reading Motivation	--	.16	.41*	--

*Note:* \*\* $p < .01$ , two-tailed. \* $p < .05$ , two-tailed.

## **Preliminary Data Analyses for Comparative Study**

**Word identification.** To ensure that participants were adequately matched on word identification ability, the groups were compared on their word identification age equivalence scores. An aggregated word identification age equivalence score was calculated from both word identification subtests and used in the present analysis. Because the data violated the assumption of normality, a Mann-Whitney U test was used in place of an independent samples *t*-test. The Mann-Whitney U test revealed no significant difference between the group with DS ( $Md = 7.25$ ,  $n = 20$ ) and the TD group ( $Md = 7.40$ ,  $n = 20$ ) on word identification,  $U = 186.50$ ,  $z = -.37$ ,  $p = .72$ ,  $r = .06$ . Because the *p* value was greater than .5, groups were considered equivalent (see Mervis and Robinson, 2003) in word identification ability, and later analyses did not control for word identification.

**Mental age.** Group comparisons were also made to see how groups compared on verbal mental age, using KBIT-2 verbal age equivalence scores, and visual analysis skill, using the KBIT-2 nonverbal age equivalence scores. For both tests, the data violated the assumption of normality, and Mann-Whitney U tests were used in place of independent samples *t*-tests to compare groups. Results of the Mann-Whitney U test for verbal mental age revealed a significant difference between groups,  $U = 98.00$ ,  $z = -2.76$ ,  $p = .005$ , with a medium effect,  $r = .44$ . The TD group ( $Md = 7.38$ ,  $n = 20$ ) scored significantly higher than the group with DS ( $Md = 5.58$ ,  $n = 20$ ) on verbal mental age.

When calculating nonverbal age equivalence scores from the KBIT-2, two participants with DS scored in the “less than 4.0 years” category. These data points were set at 3.5 years to be included in data analysis. Results of the Mann-Whitney U test for visual analysis skill also revealed a significant difference between groups,  $U = 49.00$ ,  $z = -4.10$ ,  $p < .001$ , with a large

effect,  $r = .65$ . The TD group ( $Md = 28.05, n = 20$ ) again scored significantly higher than the group with DS ( $Md = 12.95, n = 20$ ).

**Receptive vocabulary.** To see how the groups compared on receptive vocabulary, an independent samples  $t$ -test was used to compare groups on PPVT growth score values. All assumptions of independent samples  $t$ -tests were met. Results of the  $t$ -test indicated a significant difference between groups,  $t(38) = -2.86, p = .007$ , with a large effect, eta squared = .18. The TD group ( $M = 157.15, SD = 12.26$ ) scored significantly higher than the group with DS ( $M = 144.60, SD = 15.36$ ),

### **Primary Data Analyses for Comparative Study**

A one-way between groups multivariate analysis of variance was used to compare groups (DS vs. TD) on orthographic knowledge (dependent variables: Orthographic Choice, Orthographic Awareness, Exception Word Reading). Preliminary assumption testing revealed that Exception Word Reading was negatively skewed for both groups and violated the assumption of normality. All other assumptions of MANOVA were met. To control for this violated assumption the data were analyzed in several ways. First, a reflect and logarithm transformation was used to transform Exception Word Reading into a normal distribution, and the transformed variable was used in the MANOVA. Second, Exception Word Reading was pulled from the MANOVA and analyzed separately using a Mann-Whitney U test. Orthographic Choice and Orthographic Awareness were still analyzed using a MANOVA. Third, Exception Word Reading was analyzed as is (not normally distributed) as part of the MANOVA. All three analyses revealed the same pattern of results.

Because all three analyses revealed the same pattern of results, to avoid altering the data using a transformation and to reduce inflating the Type I error risk by using extra analyses, the

results of the MANOVA using the original data are reported. Results of the MANOVA revealed a statistically significant difference between groups on the combined dependent variables  $F(3, 36) = 6.35, p = .001$ ; Wilks' Lambda = .65; this was a large effect, partial eta squared = .35. When the results for the dependent variables were considered separately, the only difference to reach statistical significance was Orthographic Awareness,  $F(1, 38) = 5.01, p = .03$ , with a medium to large effect size, partial eta squared = .12. The TD group ( $M = 47.20, SD = 9.20$ ) performed significantly better than the group with DS ( $M = 41.30, SD = 7.34$ ). Orthographic Choice,  $F(1, 38) = .16, p = .69$ , partial eta squared = .004, and Exception Word Reading,  $F(1, 38) = .98, p = .33$ , partial eta squared = .03 were nonsignificant.

A Mann-Whitney U test was also used to compare groups on phonological recoding. The Mann-Whitney U was chosen in place of an independent samples *t*-test because the data violated the assumption of normality. Results of the Mann-Whitney U test revealed a significant difference between groups,  $U = 98.50, z = -2.76, p = .006$ , with a medium effect,  $r = .44$ . The TD group ( $Md = 8.00, n = 20$ ) scored significantly higher than the group with DS ( $Md = 3.50, n = 20$ ). See Table 8 for means, medians and standard deviations of comparative child study variables.

Table 8

*Means, Medians and Standard Deviations of Child Study Variables*

	DS n = 20			TD n = 20		
	Mean	Median	SD	Mean	Median	SD
Word Identification	7.64	7.25	1.01	7.60	7.40	1.18
KBIT-2 verbal mental age	6.14	5.58	1.74	7.48	7.38	1.38
KBIT-2 visual analysis skill	5.24	5.08	1.43	8.12	8.25	2.28
PPVT growth score value	144.60	141.50	15.36	157.15	155.50	12.26
Orthographic Choice	56.20	57.50	11.73	54.75	54.00	11.00
Orthographic Awareness	41.30	42.00	7.36	47.20	47.00	9.20
Exception Word Reading	19.40	21.00	5.37	17.60	20.00	6.09
Phonological Recoding	4.75	3.50	4.94	8.85	8.00	5.07

**Exploratory Analyses**

**Recognition vs. production.** To explore the hypothesis of an interaction between group (DS vs. TD) and measure of orthographic knowledge (recognition vs. production) an exploratory, mixed between-within subjects analysis of variance was conducted to assess group performance on three different orthographic measures (Orthographic Choice, Orthographic Awareness and Exception Word Reading). All three measures were standardized using z-scores for data comparison. Because z-scores were used in data analysis, setting the mean equal to zero for each measure, the main effect of test could not be determined.

Results of the ANOVA revealed a significant interaction between group and measure, Wilks Lambda = .66,  $F(2, 37) = 9.58$ ,  $p < .001$ , with a large effect, partial eta squared = .34. The main effect comparing groups was not significant,  $F(1, 38) = .08$ ,  $p = .79$ , partial eta squared = .002. See Figure 2 for interaction. See Table 9 for means and standard deviations.

Independent samples *t*-tests were then used to compare groups on each of the three dependent variables using z-scores. Results of the independent samples *t*-tests revealed a

significant difference between groups on Orthographic Awareness,  $t(38) = -2.24, p = .03$ , with a large effect,  $\eta^2 = .17$ . The TD group performed significantly better than the group with DS. Results did not reveal a significant difference between groups on Exception Word Reading,  $t(38) = .99, p = .33, \eta^2 = .03$  or Orthographic Choice,  $t(38) = .40, p = .69, \eta^2 = .004$ . These results are not consistent with differential group effects of recognition vs. production tasks. See Table 9 for group means and standard deviations.

Figure 2

*Recognition vs. Production Measures of Orthographic Knowledge*

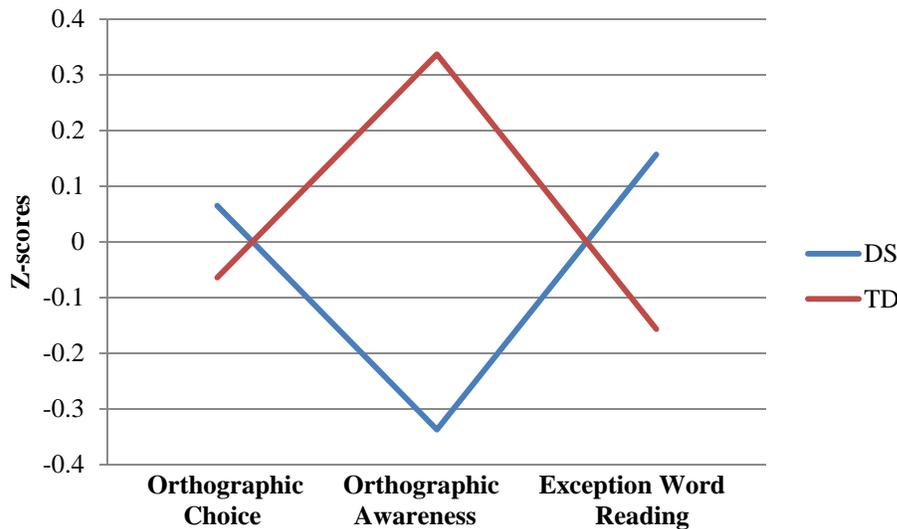


Table 9

*Means and Standard Deviations of Orthographic and Phonological Task Z-scores*

	DS <i>n</i> = 20		TD <i>n</i> = 20		z-score difference
	Mean	SD	Mean	SD	
Orthographic Choice	.06	1.044	-.06	.98	.12
Orthographic Awareness	-.34	.84	.34	1.05	.68
Exception Word Reading	.16	.94	-.16	1.06	.32
Phonological Recoding	-.38	.92	.38	.95	.76

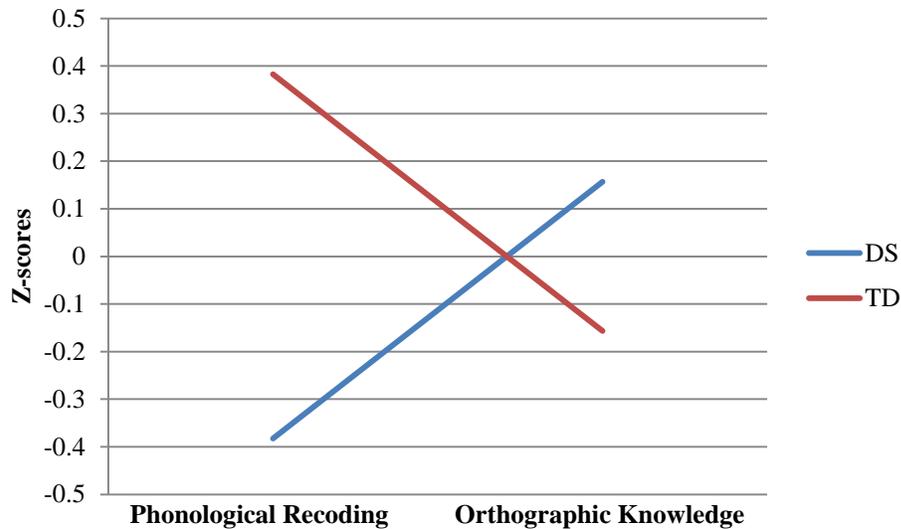
**Phonological recoding vs. orthographic knowledge.** To explore the hypothesis of an interaction between group (DS vs. TD) and subskill of word identification (phonological recoding vs. orthographic knowledge) an exploratory, mixed between-within subjects analysis of variance was conducted to assess group performance on two different reading measures (phonological recoding vs. Exception Word Reading). Word Attack was used as the measure of phonological recoding. Exception word reading was chosen as the measure of orthographic knowledge as it, consistent with the measure of phonological recoding, requires a verbal production by the participant. Phonological recoding and Exception Word Reading were standardized using z-scores for data comparisons. Because z-scores were used in data analysis, setting the mean for both tasks at zero, main effect of test could not be determined.

Results of the ANOVA revealed a significant interaction between group and measure, Wilks Lambda = .92,  $F(1, 38) = 1.12$ ,  $p = .007$ , with a large effect, partial eta squared = .18. The main effect comparing groups was not significant,  $F(1, 38) = .87$ ,  $p = .36$ , partial eta squared = .02. See Figure 3 for interaction. See Table 9 for means and standard deviations.

As noted above, results of an independent samples *t*-test did not reveal a significant difference between groups on Exception Word Reading using z-scores,  $t(38) = .99$ ,  $p = .33$ , eta squared = .03. An independent samples *t*-test was also used to compare groups on phonological recoding using z-scores. Results of the *t*-test revealed a significant difference between groups on phonological recoding,  $t(38) = -2.59$ ,  $p = .01$ , with a large effect, eta squared = .15. The TD group performed significantly better than the group with DS. See Table 9 for group means and standard deviations.

Figure 3

*Phonological Recoding vs. Orthographic Knowledge*



**Receptive vocabulary.** Pearson product-moment correlations were used to explore the relationship between receptive vocabulary and reading for each group (DS vs. TD) separately. Within the group with DS, receptive vocabulary was significantly correlated with all reading measures. In contrast, within the TD group, receptive vocabulary was only correlated with word identification. The correlations of receptive vocabulary and word identification for each group were statistically compared. Results did not reveal a significant difference between groups, as  $z = 1.10$  was not less than  $-1.96$  or greater than  $1.96$ . See Table 10 for correlations.

Table 10

*Correlations between Receptive Vocabulary, IQ and Reading Measures (correlations for the group with DS above the diagonal, correlations for the TD group below the diagonal)*

	IQ	PPVT	Word Identification	Phonological Recoding	Orthographic Choice	Orthographic Awareness	Exception Words
IQ	--	.67**	.47*	.51*	.53*	.59**	.35
PPVT	.43	--	.79**	.82**	.69**	.54*	.63*
Word Identification	.15	.60**	--	.93**	.66**	.59*	.76**
Phonological Recoding	-.22	.10	.18	--	.64**	.57**	.70**
Orthographic Choice	-.19	.38	.82**	.10	--	.68**	.55*
Orthographic Awareness	-.21	.36	.75**	.04	.84**	--	.40
Exception Words	-.08	.40	.76**	-.14	.87**	.86**	--

Note: \*\* $p < .01$ , two-tailed. \* $p < .05$ , two-tailed.

**IQ.** Pearson product-moment correlations were also used to explore the relationship between IQ and reading for each group (DS vs. TD) separately. Within the group with DS, IQ was significantly correlated with almost all reading measures. Exception Word Reading was the only reading variable not significantly correlated with IQ in DS. In contrast, within the TD group, IQ was not significantly correlated with any reading measures. See Table 10 for correlations.

**Parent questionnaire and children's data.** Within the DS sample, correlations between parent reports of child's reading (including reading ability, the home literacy environment and child's motivation to read), the child's tested reading abilities (including word identification, phonological recoding, and orthographic knowledge), and the child's receptive vocabulary were examined for exploratory purposes. From the parent survey, sum scores from three subscales (reading ability, home literacy environment and child's reading motivation as reported by the parent) were used in the present analyses. For the reading ability subscale, a sum score was

calculated using 10 of the 11 questions. The first question (“Can your child recite the alphabet?”) was excluded because one parent had left this question blank, but due to the correlational design, the investigators wished to maximize the number of participants included. See Table 11 for correlations.

Table 11

*Correlations between Parent Survey and Comparative Child Studies (corrected for age below the diagonal)*

	Age	RA	RM (P)	HLE	RM (C)	WID	PR	OC	OA	EWR	PPVT
Age	--	.30	.50*	-.37	.00	.25	.26	.53*	.13	.17	.21
RA	--	--	.07	.16	-.30	.55*	.57**	.66**	.34	.55*	.76**
RM (P)	--	.46	--	.64**	.09	.16	.23	-.02	.12	.13	.22
HLE	--	.52*	.58*	--	.20	.47*	.52*	.20	.31	.34	.66**
RM (C)	--	-.30	.10	.21	--	-.09	-.09	-.36	-.27	-.16	-.04
WID	--	.54*	.32	.61**	-.10	--	.93**	.66**	.56*	.76**	.79**
PR	--	.55*	.41	.68**	-.09	.93**	--	.64**	.57**	.70**	.82**
OC	--	.62**	.29	.48*	-.42	.64**	.62**	--	.68**	.55*	.69**
OA	--	.30	.19	.39	-.27	.56*	.56*	.73**	--	.40	.54*
EWR	--	.55*	.23	.43	-.17	.75**	.69**	.56*	.39	--	.63**
PPVT	--	.75**	.36	.80**	-.05	.77**	.81**	.70**	.54*	.62**	--

*Note:* RA = reading ability. RM (P) = reading motivation reported by parent. HLE = home literacy environment. RM(C) = reading motivation reported by child. WID = word identification. PR = phonological recoding. OC = Orthographic Choice. OA = Orthographic Awareness. EWR = Exception Word Reading.

\*\* $p < .01$ , two-tailed. \* $p < .05$ , two-tailed.

## DISCUSSION

The primary goal of this dissertation was to measure orthographic knowledge, the visual subskill of word identification, in individuals with DS. It examined the word identification domain of the Simple View of Reading (Gough & Tunmer, 1986) by matching participants with DS to TD participants on word identification ability. It then measured two important subskills of word identification: phonological recoding and orthographic knowledge. Based on previous research, it was expected that participants with DS would perform worse on phonological recoding. However, it was hypothesized that individuals with DS would perform better on measures of orthographic knowledge by capitalizing on their strength in visuospatial processing.

A secondary goal of this dissertation was to estimate literacy rates in a DS registry sample. Previous studies have varied in their reports of the number of participants with DS who can and cannot read. The present study addressed this issue by surveying a large sample of parents of children with DS about their children's reading skills. This also allowed me to select a sample of individuals with DS reported to have stronger reading skills to participate in the comparative child study. By using this two-part approach, the results of the comparative child study can be placed in a larger context, which previous studies have not done. The parent survey also allowed me to measure non-cognitive factors, such as the home literacy environment, to see how these impact reading development in DS.

### **Word Identification**

In the present study the average word identification ability for both groups was approximately 7 years. Because the *p* value was greater than .5 (see Mervis and Robinson,

2003), I concluded that the groups were equivalent in terms of word identification ability and did not statistically control for this in the remaining analyses. Matching on word identification allowed me to then examine strengths and weaknesses of the subskills of word identification in DS. When matched on word identification, the group with DS performed worse on measures of verbal mental age, visual analysis skill and receptive vocabulary. These findings are consistent with previous research showing similar patterns (Cupples & Iacono, 2000; Fowler et al., 1995; Kay-Raining Bird et al., 2000) and indicate that word identification ability surpasses what would be expected based on verbal mental age, visual analysis skill and vocabulary in individuals with DS.

As noted in the introduction, matching is a common concern in research on DS. Matching on one variable always causes mismatches on additional variables other than the dependent variable. The present study illustrates this issue perfectly. By matching on word identification, participants were then mismatched on chronological age, IQ, verbal mental age, visual analysis skill and receptive vocabulary. While not perfect, a word identification match was the best suited method for the present study as it allowed for a better understanding of its subskills (phonological recoding and orthographic knowledge) than other matches would have provided.

### **Orthographic Knowledge and Phonological Recoding**

The primary goal of this study was to measure orthographic knowledge in individuals with DS. I hypothesized that individuals with DS would perform better overall on orthographic knowledge when recognition measures were included in the protocol. This hypothesis was not supported. Whereas participants with DS performed slightly better than the TD group on two of the orthographic tasks, they performed significantly worse on a third orthographic task.

However, overall participants with DS appear to have orthographic knowledge that is consistent with their word identification level. This finding is consistent with previous research involving production-only measures (Cardoso-Martins et al., 2009; Gombert, 2002; Roch & Jarrold, 2008; Verucci et al., 2006).

An interaction between group and type of orthographic task (recognition vs. production) was also hypothesized, with the group with DS performing better on recognition measures than a production measure. This hypothesis was not supported either. The group with DS did not perform significantly different than the TD group on Orthographic Choice, which was a recognition measure, or on Exception Word Reading, which was a production measure. Further, the group with DS showed different patterns for the two recognition measures. Whereas they did not perform significantly different than the TD group on Orthographic Choice, they performed significantly worse on Orthographic Awareness.

Although the original interaction hypothesis was not supported, a significant interaction between group and orthographic task did emerge. The TD group performed significantly better on the Orthographic Awareness task whereas the group with DS performed somewhat better on both Orthographic Choice and Exception Word Reading. This pattern does not suggest a recognition vs. production distinction in orthographic knowledge for individuals with DS. Rather, it suggests a real word vs. nonword distinction. Both Orthographic Choice and Exception Word Reading used real words; Orthographic Awareness used letter patterns, not real words.

Although research is scarce, the reading community is beginning to recognize these different components of orthographic knowledge. Apel (2011) proposed a model of orthographic knowledge with two broad subtypes: *word-specific* and *general* (referred to by Apel

as mental graphemic representations and orthographic rules, respectively). Word-specific orthographic knowledge relates to actual words that children have learned to identify. General orthographic knowledge relates to sensitivity to letter combinations that are legal and probable but not necessarily to actual words. There are several possible reasons why individuals with DS may show stronger word-specific orthographic knowledge and weaker general orthographic knowledge.

One possible explanation is the role of experience in orthographic knowledge. Burt (2006) suggested that word-specific orthographic knowledge is influenced more by reading experience than general orthographic knowledge. The participants with DS in this study were older and likely had more experience with print and reading than did the TD participants. Because individuals with DS may require more repetition to acquire the same level of reading, it makes sense that they would need more years of experience to acquire the same level of word-specific orthographic knowledge.

It also makes sense that with more years of experience they would have built up stronger word-specific orthographic knowledge, but not necessarily stronger general orthographic knowledge. If reading experience influences word-specific orthographic knowledge, then it is possible that accumulated word-specific orthographic knowledge influences general orthographic knowledge in the typical population. The more real words an individual encounters, the greater sensitivity they may develop to word and letter patterns found in their language. The poorer performance in general orthographic knowledge by individuals with DS, as measured by the Orthographic Awareness task, suggests that these individuals may have difficulty generalizing common letter patterns from real words.

Another possible explanation for the group difference in Orthographic Awareness is that the task was too difficult. A more in-depth analysis of this particular task revealed that 7 of 20 participants with DS and four of the TD participants performed at chance level on this task (set at 55% accuracy or below). However, the task could be designed to be easier (e.g., Allington, 1981) or more difficult. Results of this study suggest that for a given word identification level, individuals with DS performed worse on the Orthographic Awareness task than would be expected. More research is needed on general orthographic knowledge in DS.

As hypothesized, and consistent with previous research, participants with DS performed worse on phonological recoding than did the TD participants. Also as hypothesized, there was a significant interaction between groups in phonological recoding and orthographic knowledge. The TD group performed significantly better on phonological recoding whereas the group with DS performed slightly better on Exception Word Reading. These results suggest that while individuals with DS struggle with phonological recoding, orthographic knowledge is congruous with word identification level. This finding is also consistent with the above-mentioned results of the recognition vs. production measures of orthographic knowledge. In this analysis both the phonological recoding and orthographic knowledge tasks required verbal production by participants. Participants with DS performed significantly worse on one (Word Attack) but not the other (Exception Word Reading). This indicates that requiring a verbal production response is not to blame for poor performance in phonological recoding.

When matched on word identification ability, the participants with DS performed worse on phonological recoding, but similarly on orthographic knowledge. This raises the question of how participants with DS are achieving the same level of word identification ability as younger TD children with higher mental ages and better phonological recoding skills. First, it is possible

that individuals with DS are relying more heavily on orthographic knowledge than TD children to read words. TD children can utilize both phonological recoding and orthographic knowledge to read words, but with such poor phonological recoding abilities, individuals with DS may rely more on acquired orthographic knowledge to read words.

This idea fits with connectionist theories (Seidenberg, 2005; Seidenberg & McClelland, 1989; Plaut & McClelland, 2000; Plaut, McClelland, Seidenberg, & Patterson, 1996) of word identification. These developmental theories focus on how individuals master word reading. According to connectionist theories, the ability to recognize and pronounce words involves statistical learning. Networks of orthography, phonology and semantics are represented on a continuum, the strength of which is determined by connections between networks. Connections are established when an input group (i.e. orthography) activates an output unit (i.e. phonology). These connections establish important patterns of activity, such as a phonological pathway between orthographic and phonological groups and a semantic pathway between orthographic and semantic groups. According to connectionist models, poor language abilities in individuals with DS may lead to problems developing a phonological pathway, causing impaired phonological recoding, as seen in the present study. On the other hand, stronger visual skills could lead to a relatively stronger orthographic pathway, leading to relatively stronger orthographic knowledge, as seen in the present study. In this model, individuals with DS may regularly utilize their stronger orthographic pathway to read words.

A second explanation for how individuals with DS acquire the same level of word identification as TD children despite poorer phonological recoding and similar orthographic knowledge is that these individuals, with more reading experience, have memorized a larger number of real words than TD children. This idea fits with dual-route theories of word

identification (Coltheart, 1978; 1980; 1996; 2000; Coltheart et al., 1993; Coltheart et al., 2001; Forster & Chambers, 1973). These models include two routes: lexical and nonlexical. The lexical route involves an individual looking up a familiar word stored in his/her lexicon. The nonlexical route involves phonologically recoding new and unfamiliar words when reading. Once unfamiliar words become familiar, they are then stored in the lexicon, and an individual can access them via the lexical route while reading. According to this model, individuals with DS may have acquired a larger lexicon of real words from which to draw upon when reading. This would explain how individuals with DS read as many words as TD children despite having worse phonological recoding abilities and similar levels of orthographic knowledge. Further, each word in the lexicon has a meaning, and when individuals access words they also access the meanings of those words. This could explain Buckley's (1985) finding that individuals with DS make more semantic errors than phonological errors when reading.

### **Receptive Vocabulary and IQ**

Based on previous research showing a relationship between receptive vocabulary and word identification in DS (Boudreau, 2002), the present study hypothesized a significant relationship between receptive vocabulary and reading, including orthographic knowledge. This hypothesis was clearly seen in the group with DS, as PPVT scores showed significant, positive correlations with word identification, phonological recoding, Orthographic Choice, Orthographic Awareness and Exception Word Reading. In contrast, PPVT scores only correlated with word identification in the TD sample. Overall, receptive vocabulary was more strongly related to reading in the group with DS as it correlated with a larger number of reading measures. However, the correlations between receptive vocabulary and word identification were not

significantly different between the two groups, suggesting that stronger vocabularies are associated with better word identification to a similar degree in both groups.

Interestingly, within the TD group, phonological recoding was not significantly correlated with any of the other reading measures. The relationship between phonological recoding and word identification within TD participants is well-established in the literature (e.g., Connors, 2009; Cunningham et al., 2001; Manis et al., 1999; Torgesen et al., 1997). The lack of a significant correlation in this study was most likely due to sampling. While the data were normally distributed, some younger participants scored very high on phonological recoding despite lower word identification scores, and some older TD participants scored high on word identification despite lower phonological recoding scores. I believe with a larger sample, these data would wash out and a significant correlation would emerge.

One concern in conducting research with any ID sample, DS included, is IQ. Research on the relationship between reading and IQ is mixed. Some research with both TD children and children with DS has suggested that intelligence is related to reading ability (Cardoso-Martins et al., 2009; Carr, 1995; Ellis & Large, 1988; Sloper et al., 1990; Stanovich et al., 1984). Other research with participants with ID has failed to find significant relationships between reading and IQ (Connors, 1990; Slate, 1995). In the present study IQ was significantly correlated with reading in DS, but not in TD. All TD participants had IQs within a normal range (83-125), whereas the participants with DS had IQs ranging from 40-66. It appears that for TD children with normal IQs, slightly higher IQs do not significantly aid in reading. However, for individuals with DS with IQs in the ID range, higher IQs can and do aid in reading. The present study chose to focus on word identification ability and not IQ, but future studies may be interested in further researching the relation between IQ and reading.

## **Parent Survey**

One goal of the present study was to estimate literacy rates in a registry sample of individuals with DS. Out of those surveyed with children over the age of five, 95.9% of parents reported that their child could read single words and 70.8% reported that their child could sound out new or unfamiliar words. Finally, 73% of parents reported that they would describe their child as a reader. Taken together, these reports suggest that a majority of individuals with DS have some reading ability. Almost all individuals with DS can read single words, and many individuals with DS also have some phonological recoding ability and can sound out new or unfamiliar words. However, whereas 66.7% of parents reported their child could read single words very well, only 25% reported that their child could sound out new/unfamiliar words very well. Word identification appears to be a strength for individuals with DS relative to general cognitive ability, and phonological recoding appears to be impaired relative to word identification.

Though limited, previous research has reported fairly rich home literacy environments for individuals with DS (Ricci, 2011). The present study also reflected rich home literacy environments for individuals with DS. Almost all parents reported that they began reading to their child before their child was one year old. They also reported reading to their child frequently and always having children's books in the home.

Previous research with TD children has indicated that rich home literacy environments are linked to increases in children's interest in reading (Lyytinen et al., 1998). In her study, Ricci (2011) also reported that the grade-school children with DS with richer home literacy environments indicated more interest in reading than pre-school age children with DS with poorer home literacy environments. The present study was consistent with both Lyytinen et al.,

(1998) and Ricci's (2011), findings that the home literacy environment is significantly correlated with parent reports of the child's motivation to read. Although not directly tested, rich home literacy environments when the child is very young may lead to increases in the child's motivation to read. One unexpected, negative significant correlation emerged between age and reading motivation. This correlation can most likely be explained by the specific questions asked in the reading motivation portion of the parent survey. Two of the four reading motivation questions asked about child interest in reading to or with parents. These results indicate that as children age, they become less interested in reading with their parent.

In TD children, exposure to, and experience of, rich home literacy environments has been found to contribute to later reading achievement and language abilities (Bus et al., 1995; Lyytinen et al., 1998; Scarborough & Dobrich, 1994; Senechal, 1997). The present study found a significant, medium, positive correlation between home literacy environment and receptive vocabulary. The present study did not find a significant correlation between home literacy environment and parent reports of the child's reading ability. However, the home literacy environment was significantly correlated with reading abilities measured in the comparative child study. The home literacy environment, as reported by parents, was moderately and significantly correlated with word identification and phonological recoding. Home literacy environment was not significantly correlated with the orthographic tasks. Despite lack of significance, correlations between the home literacy environment and Orthographic Awareness and Exception Word Reading were .31 and .34 respectively. It is possible that with a larger sample size these correlations could reach conventional levels of significance. Though not directly tested, it is possible that rich home literacy environments lead to more exposure to reading and better reading abilities.

Despite significant differences in the correlations between home literacy environment and parent reports of child's reading abilities (nonsignificant) and child's actual reading abilities (significant), parents seemed to be fairly accurate in reporting their child's reading abilities. Parent reports of their child's reading abilities were significantly correlated with word identification, phonological recoding, Orthographic Choice and Exception Word Reading.

### **Limitations and Future Directions**

One limitation of the parent survey is a possible ascertainment bias. This bias could be associated with the UAIDPR and/or with the set of parents from the UAIDPR who wished to participate in the study. It is possible that parents and families who join the UAIDPR only represent a subset of the population of families who have a child with DS. For example, families who join the UAIDPR could possibly have higher social economic status, be more interested in research and education and have greater access to resources for their child with DS than families who have not joined the UAIDPR. It is also possible that those families from the UAIDPR who wished to participate in this particular study only represent a sample of the population of families who have a child with DS. For example, it is possible that parents with a child with DS who could read were interested in participating, while those whose child with DS could not read were not interested in participating.

The child study showed that individuals with DS performed as well as TD children matched on word identification on two measures of orthographic knowledge, despite weaker phonological recoding. However, this finding should be replicated in other samples of individuals with DS. The participants with DS recruited for the present study were selected because they were between the ages of 11 and 21 and were reported by their parents to be able to

read words. Had a group of participants with DS with higher or lower word identification abilities been selected, the results may have differed.

The child study consisted of a wide age range of participants with DS, adding heterogeneity to the group. A next step for researchers would be to conduct research with more limited age ranges in DS. Starting with younger children and conducting research with more limited age groups would allow researchers to more fully investigate the development of orthographic knowledge in DS. It would also be interesting to include comparison groups of mixed etiology ID to more fully explore the role of IQ in reading as well as the cognitive profile of DS.

Another limitation of the present study was the differences between groups in terms of gender and race. The group with DS was 60% female and 40% male whereas the TD group was 40% female and 60% male. Further, the TD group was much more racially diverse than the group with DS. Despite group gender differences, there were no gender differences within each group on any of the reading measures. Additionally, despite a more racially diverse TD sample, TD minority students did not score significantly differently than their Caucasian peers. However, it is unclear how additional participants with DS from racial minorities would have performed in the present study.

While consistent with other research, the present study also had a small sample size, which may have limited the power necessary to detect a significant difference between groups. With larger samples, undetected group differences from this study may emerge. Further, with larger samples, additional statistical techniques, such as developmental trajectories and hierarchical linear modeling, could be used to examine changes between groups and within groups across development.

More research is needed on orthographic processing, especially on the development of orthographic knowledge, orthographic learning, and the differences between word-specific orthographic knowledge and general orthographic knowledge. Some research has suggested that word shape facilitates reading development in TD children by acting as a visual cue (Webb, Beech, Mayall & Andrews, 2006), and future research could explore this as a possible underlying strength of word identification and orthographic knowledge in DS. Another interesting avenue of future research is to explore the relationship of experience and instruction to reading in DS, especially as it relates to orthographic knowledge. These are just a few of the next steps needed to more fully understand reading in individuals with DS.

With more research, researchers will be better equipped to design and implement reading interventions and improved reading instruction programs for individuals with DS. Whereas the individuals with DS in the present study performed as well as younger TD children on word identification, it is important to remember that word identification is only a relative strength in DS. The participants with DS in the present study were reading well-below their chronological age. However, if orthographic processing is a strength in DS, then it may be possible to utilize encoding (spelling) instruction to improve phonological recoding and word identification skills. Likewise, increased instruction in phonological recoding could lead to even better orthographic skills, and together these skills could lead to improving word identification abilities and ultimately reading comprehension.

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## LIST OF APPENDICES

A. Exception Words and Kuchera & Francis (1976) Frequency Ratings.....	84
B. Parent Reading Questionnaire.....	85
C. Child Reading Motivation Questionnaire.....	89
D. Institutional Review Board Approval – Parent Study.....	90
E. Institutional Review Board Approval – Child Study.....	91

## Appendix A

### Exception Words and Kuchera and Francis (1976) frequency ratings

1. Come - 630
2. Give - 391
3. Own - 772
4. Great - 665
5. Lose – 58
6. Soul – 47
7. Good - 807
8. World - 787
9. Island - 167
10. Bowl – 23
11. Friend -133
12. Eye - 122
13. Sure - 264
14. Ceiling – 31
15. Four -359
16. Answer - 152
17. Blood - 121
18. Are - 4393
19. Break – 88
20. Said – 1961
21. Do – 1363
22. Have – 3941
23. Both – 730
24. What – 1908
25. Move - 171

Appendix B

Parent Reading Questionnaire

**Instructions:** The primary caregiver should fill out the following survey. If a question does not apply please write "N/A" for not applicable. If you are not sure, please write "not sure".

Your relation to the child: \_\_\_\_\_

Child's birthdate: \_\_\_\_\_ Child's age: \_\_\_\_\_

Child's gender (please circle):      Male    Female

What is your highest level of education?

\_\_\_ some high school

\_\_\_ high school diploma

\_\_\_ some college or vocational training

\_\_\_ college degree

\_\_\_ graduate degree

**Hearing & Vision**

1. Does your child have any vision impairments (with or without glasses)?      \_\_\_ Yes      \_\_\_ No

If yes, how old was your child when you first realized he/she had vision impairment? \_\_\_\_\_

2. Does your child wear corrective lenses or glasses?      \_\_\_ Yes      \_\_\_ No

If yes, how old was your child when he/she received corrective lenses or glasses? \_\_\_\_\_

3. Does your child have any hearing loss?      \_\_\_ Yes      \_\_\_ No

If yes, how old was your child when you first realized he/she had hearing loss? \_\_\_\_\_

4. Does your child wear a hearing aid?      \_\_\_ Yes      \_\_\_ No

If yes, how old was your child when he/she received the hearing aid? \_\_\_\_\_

**Language**

1. Does your child use language as his/her main way to communicate? \_\_\_ Yes \_\_\_ No

2. How would you rate your child's overall language ability compared to that of other children with Down syndrome? (Please circle your response).

1	2	3	4
No language	Poor	Average	Strong

**Reading abilities** – Please circle your response.

1. Can your child recite the alphabet?

1	2	3
No, not at all	Somewhat	Yes, very well

2. Can your child identify single, written letters?

1	2	3
No, not at all	Somewhat	Yes, very well

3. Can your child write out letters?

1	2	3
No, not at all	Somewhat	Yes, very well

4. Does your child know sounds of letters?

1	2	3
No, not at all	Somewhat	Yes, very well

5. Can your child read single words?

1	2	3
No, not at all	Somewhat	Yes, very well

6. Can your child sound out new/unfamiliar words?

1	2	3
No, not at all	Somewhat	Yes, very well

7. Does your child read basic picture books or simple stories?

1	2	3
No, not at all	Somewhat	Yes, very well

8. Does your child read books that contain new words or concepts?

1	2	3
No, not at all	Somewhat	Yes, very well

9. Does your child read magazines?

1	2	3
No, not at all	Somewhat	Yes, very well

**10. Does your child read news stories?**

1	2	3
No, not at all	Somewhat	Yes, very well

**11. Would you describe your child as a reader?**

1	2	3
No, not at all	Somewhat	Yes, very well

**Reading Motivation** – Please circle your response. If you are unsure, write in “Not sure”.

**1. In a typical week, how often does your child amuse him/herself with books, magazines or comic books?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**2. In a typical week, how often does your child ask you to read to him/her?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**3. How interested is your child in reading books with you?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**4. How interested is your child in other reading activities?**

1	2	3
No interest	A little interest	A lot of interest

**Home Literacy Environment** – Please circle your answer. If a question does not apply, please write “N/A” for not applicable. If you are unsure, please write “not sure”.

**1. Approximately how many children’s books did/do you have in the home:**

**a. When your child was an infant (birth to 1 year of age)?**

1	2	3	4	5
None	0-50 books	50-100 books	100-200 books	More than 200 books

**b. When your child was a toddler (2 to 3 years of age)?**

1	2	3	4	5
None	0-50 Books	50-100 Books	100-200 Books	More than 200 Books

**c. Approximately how many children’s books are currently in your home?**

1	2	3	4	5
None	0-50 Books	50-100 Books	100-200 Books	More than 200 Books

**2. Approximately how many adult-level books are in your home?**

1	2	3	4	5
None	0-50 Books	50-100 Books	100-200 Books	More than 200 Books

**3. At what age did you begin reading to your child?**

1	2	3	4	5	6	7
Not yet	Birth to 6 months	7 months to 11 months	1 year to 2 years	3 years to 4 years	5 years to 6 years	7 years or older

**4. How often did you read to or with your child, including books, magazines, stories on e-readers, comic books, etc.:**

**a. When he/she was an infant, birth to 1 year of age?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**b. When he/she was a toddler, 2 to 3 years of age?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**c. How often do you currently read to/with your child?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**5. How often did/do you visit the library and/or bookstore with your child:**

**a. When he/she was an infant, birth to 1 year of age?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**b. When he/she was a toddler, 2 to 3 years of age?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**c. Currently?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**6. How often do you read at home?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

**7. How often is your child aware of you or another family member reading?**

1	2	3	4	5
Never/ almost never	Weekly	A few times a week	Daily	A few times a day

## Appendix C

### Child Reading Motivation Questionnaire

Do you like reading?            Yes / No

If yes: How much do you like reading?    A little / A lot

Do you like reading books with your mom/dad? Yes / No

If yes: How much do you child like reading books with your mom/dad? A little / A lot

Appendix D

Institutional Review Board Approval – Parent Study

September 18, 2012



Susan Loveall  
Department of Psychology  
College of Arts & Sciences  
Box 870348

Re: IRB # 12-OR-313: "Reading in Down Syndrome: Parent Survey"

Dear Ms. Loveall,

The University of Alabama Institutional Review Board has granted approval for your proposed research.

Your application has been given expedited approval according to 45 CFR part 46. You have also been granted a waiver of written documentation of informed consent (telephone respondents). Approval has been given under expedited review category 7 as outlined below:

*(7) Research on individual or group characteristics or behavior (including, but not limited to, research on perception, cognition, motivation, identity, language, communication, cultural beliefs or practices, and social behavior) or research employing survey, interview, oral history, focus group, program evaluation, human factors evaluation, or quality assurance methodologies.*

Your application will expire on September 17, 2013. If the study continues beyond that date, you must complete the IRB Renewal Application. If you modify the application, please complete the Modification of an Approved Protocol form. Changes in this study cannot be initiated without IRB approval, except when necessary to eliminate apparent immediate hazards to participants. When the study closes, please complete the Request for Study Closure (Investigator) form.

**Please use reproductions of the IRB-stamped consent forms.**

Should you need to submit any further correspondence regarding this application, please include the assigned IRB application number.

Good luck with your research.

Sincerely,

Appendix E

Institutional Review Board Approval – Child Study

Office for Research  
Institutional Review Board for the  
Protection of Human Subjects



October 9, 2012

Susan Loveall  
Department of Psychology  
College of Arts and Sciences  
Box 870348

Re: IRB Application # 12-018 "Reading Skills in Down Syndrome:  
An Examination of Orthographic Processing Abilities"

Dear Ms. Loveall:

The University of Alabama IRB has received the revisions requested by the full board on 9/21/12. The board has reviewed the revisions and your protocol is now approved for a one-year period. Please be advised that your protocol will expire one year from the date of approval, 9/21/12.

If your research will continue beyond this date, complete the IRB Renewal Application by the 15<sup>th</sup> of the month prior to project expiration. If you need to modify the study, please submit the Modification of An Approved Protocol Form. Changes in this study cannot be initiated without IRB approval, except when necessary to eliminate apparent immediate hazards to participants. When the study closes, please complete the Request for Study Closure Form.

Should you need to submit any further correspondence regarding this proposal, please include the assigned IRB application number. Please use reproductions of the IRB approved stamped consent/assent forms to obtain consent from your participants.

Good luck with your research.

Sincerely,