Understanding emergent literacy and improving reading comprehension of preschool children with Autism Spectrum Disorder and hyperlexia

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Abstract

People with autism spectrum disorder (ASD), a neurodevelopmental disorder characterized by social communication challenges and repetitive patterns of behaviour, are known to have a number of strengths and special interests. Hyperlexia, a term coined by Silberberg and Silberberg (1967) represents both a strength and a special interest for people with ASD. Those with hyperlexia demonstrate a strength in early word reading, alongside an intense special interest in letters and words, from a very early age. However, this strength in early word reading is accompanied by challenges in reading comprehension. The current dissertation had several goals. First, to review the literature on young children with hyperlexia to gain a better understanding of the mechanisms specific to emergent literacy skills that underlie early word reading. A second goal, addressed in the first manuscript (Article 1), was to compare emergent literacy skills of preschool children with ASD and hyperlexia, and ASD without hyperlexia, to their typically developing (TD) peers. Findings from Article 1 indicated that preschoolers with ASD and hyperlexia demonstrate an alternate, non-phonological route to word reading that is unlike their TD peers. This study discusses the implications on teaching practices for teachers, clinicians and parents of young children with ASD and hyperlexia. Subsequently, the main goal addressed in the second manuscript (Article 2) was to evaluate a novel tablet-based, parent-supported, reading comprehension intervention aimed at improving the challenges in reading for meaning associated with hyperlexia from a very early age. The results of this 6-week intervention demonstrated gains in reading comprehension for the group with ASD and hyperlexia as compared to a TD group, and a group with ASD without hyperlexia. In addition, gains in receptive language skills were observed for all groups. Findings from Article 2 suggest a shift from teaching reading comprehension in the later grades to addressing these challenges at

the preschool level at the first signs of hyperlexia. This dissertation also adds to a growing body of research that emphasizes a strength-based approach to intervention for those with ASD.

Résumé

C'est bien connu que les personnes qui présentent un trouble du spectre autistique (TSA), un trouble neurodéveloppemental caractérisé par des problèmes de communication sociale et des comportements répétitifs, ont un certain nombre de forces et d'intérêts particuliers. L'hyperlexie, un terme introduit par Silberberg et Silberberg (1967), représente à la fois une force et un intérêt particulier pour les personnes ayant un TSA. Les personnes qui présentent à la fois avec un TSA et une hyperlexie démontrent une force en lecture de mots ainsi qu'un intérêt marqué pour les lettres et les mots dès un très jeune âge. Toutefois, cette force en lecture précoce des mots s'accompagne de défis en compréhension de lecture. Donc, la thèse actuelle a plusieurs objectifs. Premièrement, la thèse débute avec une revue de la littérature portant sur l'hyperlexie chez les jeunes enfants dans le but d'acquérir une meilleure compréhension des mécanismes, spécifiquement des capacités émergentes d'alphabétisation qui engendrent la lecture de mots précoce. Un deuxième objectif abordé dans le premier manuscrit (l'article 1) est d'examiner les compétences émergentes en alphabétisation chez les enfants d'âge préscolaire ayant un TSA avec ou sans hyperlexie par rapport à leurs pairs dont le développement est dit typique (DT). Les résultats de l'article 1 indiquent que les enfants d'âge préscolaire ayant un TSA et une hyperlexie semblent utiliser une voie alternative, et non phonologique pour la lecture de mots comme leurs pairs DT. Cette étude examine les implications quant aux pratiques éducatives d'enseignants, de cliniciens et de parents de jeunes enfants avec un TSA et une hyperlexie. Enfin, l'objectif final abordé dans le deuxième manuscrit (l'article 2), vise à évaluer une nouvelle intervention de compréhension de lecture qui s'effectue avec des tablettes iPad avec le soutien des parents et visant à améliorer les défis en compréhension de lecture associés à l'hyperlexie dès un jeune âge. Les résultats de cette intervention de six semaines ont démontré des gains dans la compréhension

écrite pour le groupe avec un TSA et une hyperlexie par rapport à un groupe DT et à un groupe avec un TSA sans hyperlexie. De plus, des gains au plan des compétences linguistiques réceptives ont été observés pour tous les groupes. Les résultats de l'article 2 suggèrent qu'un changement de l'enseignement de la compréhension de lecture en favorisant le niveau préscolaire dès les premiers signes d'hyperlexie au lieu du primaire serait de mise. Cette thèse s'ajoute également à un nombre croissant de recherches qui mettent l'accent sur une approche d'intervention axée sur les force des personnes ayant un TSA.

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Contribution to Original Knowledge

The overarching aim of this dissertation centred on elucidating the emergent literacy skills underlying word reading in preschoolers with ASD+HPL to establish recommended teaching practices, and to advance a strength-based reading intervention. Article 1 was the first study to examine the emergent literacy skills exclusively in a group of preschoolers with ASD+HPL. Previous studies which had examined the early precursors to reading, particularly phonological awareness and phoneme-grapheme correspondence skills, in preschoolers with ASD+HPL had only a few participants (Atkin & Lorch, 2006; Cardoso-Martins & Ribeiro da Silva, 2010). Thus, Article 1 was the first study to examine, in greater depth, the emergent literacy skills of preschoolers with ASD+HPL within a sample larger than one or two participants. Findings from Article 1 suggested that preschoolers with ASD+HPL are taking an alternate route to reading as compared to their TD peers. Conclusions drawn from Article 1 contributed to enhancing knowledge of instructional practices for preschoolers with ASD+HPL in both the home and the classroom.

The second aim of this dissertation was to evaluate a novel, early, strength-based intervention to support young children with ASD+HPL. Article 2 set out to assess an intervention that would capitalize on the strength in early word reading of preschoolers with ASD+HPL, aknowledge their underlying emergent literacy profile, and support their area of greatest challenge – reading comprehension. The results of Article 2 revealed the intervention had a significant impact on increasing reading comprehension scores for the preschoolers with ASD+HPL, as well as significantly increasing receptive language skills for all three groups (ASD+HPL, ASD-HPL and TD). Article 2 is the first to report on a sucessful, early reading and language intervention for preschoolers with ASD+HPL that also impacts preschoolers with ASD and those with TD. Thus, Article 2 contributes to the existing body of literature on the benefits of early intervention for preschoolers with ASD and TD preschoolers, as well as the growing body of literature on the benefits of strength-based intervention for young children with ASD+HPL. Additional objectives and contributions of these two studies to the field of early reading of preschoolers with ASD+HPL, as well as the results and interpretations are addressed within the entire dissertation, and specifically in Chapter 5.

Preface and Contribution of Authors

Chapter 1 was written and edited by Dianne Macdonald (DM) with feedback from Gigi Luk (GL) and Eve-Marie Quintin (EMQ)

Chapter 2 was written and edited by DM with feedback from GL and EMQ. The literature search, including the initial and final abstract searches, ratings and application of exclusion criteria were carried out independently on both dates by DM and Bianca Mercadante (BM).

Chapter 3 is composed of Manuscript 1, which is an exact replication (with the exception of Tables 4, 5 and 6 which are now embedded in the body rather than separated in the Appendix) of the article entitled, *Early Reading of Preschoolers with ASD, Both With and Without Hyperlexia, Compared to Typically Developing Preschoolers* published in the *Journal of Autism and Developmental Disorders (JADD)* in August 2020. The authors of this article include DM, GL and EMQ. DM conceived of the project. DM and EMQ planned and developed the methodology. DM analysed the data. DM interpreted the data with support from GL and EMQ. DM wrote and edited the article with feedback from GL and EMQ. All authors approved the final manuscript.

Chapter 4 is composed of Manuscript 2, which is an exact replication of the article entitled, *Early Reading Comprehension Intervention for Preschoolers with Autism Spectrum Disorder and Hyperlexia*, submitted to the *Journal of Autism and Developmental Disorders (JADD)* on November 19, 2020, that is currently under review. DM conceived of the project. DM and EMQ planned the methodology. DM analysed the data. DM interpreted the data with support from GL and EMQ. DM wrote and edited the article with feedback from GL and EMQ.

Chapter 5 was written and edited by DM with feedback from GL and EMQ.

Note: Manuscripts contained within this dissertation in Chapters 3 and 4 represent original contributions to the field of autism and hyperlexia research.

Chapter 1: Introduction

We read for pleasure, to learn about the world and sometimes, just to pass the time. All necessitate reading comprehension for the task to be of any substantial benefit. Reading for meaning is the ultimate goal of the reading process. But learning to read does not begin with learning to comprehend text. When we first learn to read, the focus is on learning to decode words. This process starts at the spoken word level. Typically developing children around three years of age begin to learn that the spoken words they hear in everyday conversation are made up of individual sounds and syllables. This skill is referred to as phonological awareness and it forms the basis of written word learning (Anthony & Francis, 2005; Ehri & McCormick, 1998; National Early Literacy Panel, 2008; Scarborough & Brady, 2002; Schuele & Boudreau, 2008). Children learn to play with the sounds in words through alliteration and rhyming games that show their acquisition of phonological awareness.

As they begin to see words in print, embarking on the word reading stage, their learning shifts to naming letters and matching the individual letters of the alphabet to their corresponding sounds. This process of letter naming, and sound-letter correspondence or grapheme-phoneme correspondence is another significant step in the word reading process. Sound-letter correspondence underlies the ability to decode words on a sound-by-sound basis which is integral to reading new words (National Early Literacy Panel, 2008). Once the ability to decode words is fluently established, there is a shift in focus from learning to read words, to reading for meaning or reading to learn. Typically, this shift happens in a school setting in the early elementary grades at the word and sentence levels, and gradually increases in complexity to include paragraphs in grades 2 or 3. Children then use their newly found skill of word reading to learn about various academic subjects like history, word problems in mathematics and science and progress on a path of lifelong learning.

Unfortunately, not all children are equally adept at word reading and reading for meaning. Some children have difficulty in decoding words, a condition that is generally referred to as dyslexia (Snowling et al., 2000) with associated varying subtypes or profiles (Tamboer et al., 2016). Children with dyslexia are challenged to become fluent word readers and thus, their reading comprehension can be compromised by inefficient word decoding (Snowling et al., 2020). Others may read efficiently but still be challenged to derive meaning from the connected texts they read, referred to as a reading comprehension disorder (Landi & Ryherd, 2017). One atypical form of reading comprehension disorder is referred to as hyperlexia. Hyperlexia is an atypical profile whereby children read words with a high degree of accuracy, yet their reading comprehension is impaired (Grigorenko et al., 2002; Newman et al., 2007).

In the case of hyperlexia comorbid with autism spectrum disorder (ASD), a neurodevelopmental disorder (American Psychiatric Association, 2013), children learn to read at an extremely early age, sometimes as young as age 15 months (Atkin & Lorch, 2006) and in the absence of any formal instruction. Children with hyperlexia and ASD have an intense interest and passion for letters and written words (Ostrolenk et al., 2018). Parents describe their children as voraciously attracted to symbolic representations like letters and words. On the surface, hyperlexia seems like a promising skill. Most parents are proud to report that their child can read words far in advance of kindergarten, where most typically developing children are just learning the alphabet. Yet, the challenges associated with hyperlexia are significant. That is, young children with hyperlexia usually do not have the associated reading comprehension that would otherwise be paired with such a high level of word reading (Grigorenko et al., 2002; Newman et al., 2007). Thus, while hyperlexia initially seems advantageous, the accompanying lack of reading comprehension means that reading is lacking in the one single purpose that we all conduct the activity for – gaining meaning, learning, improving our understanding and feeling pleasure. Herein lies the essential dichotomy of young children with ASD and hyperlexia. They have an exceptional skill in word reading but lack an equivalent ability to gain meaning from the words they read. Thus, reading is essentially reduced to a rote activity, focused on recitation and deprived of the ability to advance knowledge or pleasure, other than the pleasure derived from reading the words themselves.

Research has recently focused on strength-based approaches to learning (Mottron et al., 2009; Remington & Fairnie, 2017), and clearly children with ASD and hyperlexia have a strength in word reading (Grigorenko et al., 2002; Needleman, 1982; Newman et al., 2007; Silberberg & Silberberg, 1967). Therefore, the question is how to best use this strength to improve areas of challenge, namely reading comprehension? The first step is to examine their word reading process to understand whether these children learn to read like typically developing children or rather take an alternate route. Developing a greater understanding of this process might provide insight into how these children learn and afford an opportunity to support the development of meaning from the reading process.

As previously noted, typically developing children learn to read using phonological awareness and alphabet knowledge (sound-letter correspondence skills) to blend letters and sounds together to form words. Yet it is unclear whether children with ASD and hyperlexia are implementing this same approach or adopting a different means to read words. The literature surrounding this topic is conflicted, with some studies suggesting that they do possess these prerequisite skills (Westerveld et al., 2018) and others maintaining that they do not (Knight et al., 2019; Nally et al., 2018).

Thus, the first step in this dissertation is to develop an understanding of emergent literacy skills through a comprehensive literature review presented in Chapter 2. The synthesis of the literature leads to a hypothesis that children with ASD and hyperlexia may be using an alternate, non-phonological pathway to reading; however, this hypothesis was tentative given discrepancy across studies and few studies in this domain exclusively with preschoolers. Chapter 3 focuses on assessing the emergent literacy skills of preschool children with ASD and hyperlexia, children with ASD without hyperlexia, as compared to typically developing children. Findings of no significant difference between the three groups of preschoolers in phonological awareness and phoneme-grapheme correspondence, despite advanced word reading in the group with ASD and hyperlexia, suggest that children with ASD and hyperlexia are using an alternate, non-phonological approach to reading.

Finally, with a better understanding of their early word reading profile, this information was implemented in the design of an early reading comprehension intervention which sought to improve reading comprehension and oral language skills simultaneously in preschool children with ASD and hyperlexia. This intervention study is reported in Chapter 4. Results of the reading comprehension intervention study demonstrated significant increases in reading comprehension scores for preschoolers with ASD and hyperlexia as compared to the other groups. Additionally, all groups showed an increase in receptive language scores. Thus, the focus of Chapter 5 expands upon the novel contributions of these studies, and highlights the educational, clinical and professional implications of targeting reading comprehension at an earlier age, (i.e., at the first signs of hyperlexia), rather than waiting until after grade 2, when a child has failed to meet the competency level for making meaning of longer texts (Ministry of Education, Ontario, 2006; Québec Ministère de l'Éducation, 2002). Early reading and oral language comprehension intervention may be key to mitigating the academic (Ricketts et al., 2013), social (American Psychiatric Association, 2013), and language challenges (Carlsson et al., 2013) associated with ASD that negatively impact graduation rates, employment and career opportunities post-high school. This reading and oral language comprehension intervention is the first to address these challenges in children with ASD and hyperlexia at the preschool level.

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Chapter 2: Literature Review

Introduction

Hyperlexia (HPL) is a reading comprehension disorder characterized by a strength in early, superior word reading in the presence of poor reading comprehension. While hyperlexia is comorbid with a number of atypical disorders, it is most commonly associated with Autism Spectrum Disorder (ASD; Ostrolenk et al., 2017). Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder with defining features of both impaired social communication skills and repetitive patterns of behaviour as per the DSM-5 (American Psychiatric Association, 2013). The prevalence of ASD has been gradually increasing over time, but is reportedly difficult to track due to, among other factors, the heterogeneity in symptomology present in this population and the shifting diagnostic criteria over time (Baio et al., 2018). Current estimates are of approximately 1 out of 54 children are diagnosed with ASD (Maenner et al, 2020). Early diagnostic criteria for ASD in the Diagnostic and Statistical Manual of Mental Disorders (DSM) 3rd edition (DSM-III; American Psychiatric Association, 1980) included “gross deficits in language development”, whereas the DSM-IV (American Psychiatric Association, 1994) no longer included this severe language impairment requirement in the diagnostic criteria. As approximately 50% of people with ASD have a language disorder (Carlsson et al., 2013), this adjustment in diagnostic criteria was clearly an appropriate modification.

Analogous to the evolving diagnostic criteria for ASD, defining HPL has also been challenging. A recent systematic review revealed that approximately 6%-20% of children with ASD exhibit strengths in early word reading or hyperlexia (HPL; Ostrolenk et al., 2017). This range in prevalence reflects an absence of a universally accepted definition of hyperlexia (HPL). Across the literature, ascertainment of hyperlexia has been based on an indeterminant number of criteria (Ostrolenk et al., 2017). That is, more rigorous criteria as proposed by Needleman (1982), who defined hyperlexia according to five characteristics that include a very early manifestation, a developmental disorder, reading in advance of cognitive and linguistic abilities, a linguistic impairment and minimal comprehension of written material, are associated with a lower prevalence rate. In contrast, other researchers have failed to use criteria-based definitions grounded in standard deviations or age equivalence discrepancies (Grigorenko et al., 2002), or ascertained hyperlexia based on documented reports of early word reading and an associated diagnosis of ASD (Newman et al., 2007) resulting in higher prevalence rates (Ostrolenk et al., 2017). Thus, the criteria used to determine eligibility in a group characterized as having both hyperlexia and ASD varies considerably across studies.

Furthermore, even within groups of children with ASD and hyperlexia (ASD+HPL) there is much reported variation (Lin, 2013). For example, it has been proposed that an associated language disorder (Needleman, 1982) contributes to a discrepancy between language skills and reading ability, yet language skills have been reported in the average range in a group of children with ASD+HPL (Newman et al., 2007). Discrepancies such as these render the task of accurately diagnosing and developing suitable interventions extremely challenging. Consensus on the underlying word reading profile associated with ASD+HPL would permit the development of evidence-based treatment practices which, to date, are nonexistent. Therefore, the first step in developing these practices is to clarify our understanding of the linguistic basis which underlies this strength in word reading in preschool children with ASD+HPL. To do so requires clarification of the cross-study variation within the domain of ASD+HPL, specifically in studies focused on the preschool level prior to any formal exposure to early literacy instruction. To date, no reviews of emerging literacy skills of preschoolers with ASD+HPL have been conducted.

However, a recent systematic review has examined emergent literacy in preschoolers with ASD without hyperlexia (ASD-HPL; Westerveld et al., 2016). Although there were only three studies included in this review, the results also point to the heterogeneous nature of preschoolers with autism across emergent literacy skills, as well as challenges with study design (Westerveld et al., 2016). Consequently, a review the literature on preschoolers with ASD+HPL is proposed to summarize findings and to identify what factors might be contributing to the observed variation across early literacy skills. The ultimate goal is to inform our understanding of this early reading skill to better serve early educational needs in this population and support future research in this field.

Phonological versus Orthographic Decoding

Current research is conflicted as to which avenue best explains early literacy development: a phonological/sublexical route or an orthographic/lexical route, based on the dual-route hypothesis (Coltheart et al., 2001). A phonological or sublexical route centres on a sound-by-sound word reading process (sublexical), while an orthographic or lexical route focuses on whole-word recognition (sight word reading). Support for both routes relies heavily on findings of acquisition of emergent literacy skills such as, phonological awareness, phonemic awareness, nonword reading, and alphabet knowledge. That is, acquisition of these emergent literacy skills suggests a phonological or sublexical (sound-by-sound decoding) pathway to word reading whereas a lack of acquisition implies a more orthographic or lexical (whole word) approach to word reading.

In general, phonological awareness is a metalinguistic ability (Schuele & Boudreau, 2008). More specifically, it refers to “the broad class of skills that involve attending to, thinking about, and intentionally manipulating the phonological aspects of spoken language, especially the internal phonological structure of words." (Scarborough & Brady, 2002, p. 312). The phonological structure is essentially the speech sound-based structure of a word. In typically developing children (TD), phonological awareness tasks, like rhyming and segmenting words into larger chunks like syllables, are acquired prior to phonemic awareness, which involves the manipulation of smaller units, like sounds or phonemes (Carroll et al., 2003; Schuele & Boudreau, 2008). Both phonological awareness and phonemic awareness have been shown to be essential precursors to developing literacy skills in TD children (Anthony & Francis, 2005; Ehri, 2009; National Early Literacy Panel, 2008; Schuele & Boudreau, 2008). In preschoolers with ASD (without hyperlexia), studies have revealed mixed findings for the acquisition of phonological awareness. That is some studies have found preschoolers with ASD have weak phonological awareness (Knight et al., 2019; Nally et al., 2018) as compared to their TD peers (Dynia et al., 2018; Smith Gabig, 2010) while other studies have found no significant difference between TD groups and those with ASD (Westerveld et al., 2018).

Similarly, there are mixed findings for alphabet knowledge. While phonological awareness operates at the auditory level of speech sounds, alphabet knowledge is focused on the orthographic level or written letters. Both letter naming (e.g., identifying the name of the letter) and phoneme-grapheme correspondence (e.g., identifying the sound associated with the letter, also referred to as sound-letter correspondence) have been combined under one category as alphabet knowledge (National Early Literacy Panel, 2008). Developing alphabet knowledge provides support for regular word reading, referred to as word decoding. Decoding is "the process of applying one's knowledge of the correspondences between graphemes and phonemes to determine the pronunciation, and hence, the identity of the word represented by a particular letter sequence." (Scarborough & Brady, 2002, p. 324).

In preschool and kindergarten TD children, alphabet knowledge (both letter naming and phoneme-grapheme correspondence) is a strong predictor of decoding skills while phonological awareness skills are a moderately strong predictor of decoding (National Early Literacy Panel, 2008). Indeed, both alphabet knowledge and phonological awareness skills have been identified as weaknesses in reading impairments such as dyslexia and specific language impairments (Snowling et al., 2000). In contrast, for preschoolers with ASD, alphabet knowledge skills may be acquired at different rates. That is, letter naming has been found to be a strength (Dynia et al., 2017; Knight et al., 2019; Westerveld et al., 2017) yet phoneme-grapheme correspondence skills have been found to be a weakness (Knight et al., 2019; Nally et al., 2018), especially for those with poorer reading skills (Westerveld et al., 2018). In sum, it is uncertain whether preschoolers with ASD have strong or weak phonological awareness and what components of alphabet knowledge are more or less challenging.

Phonological awareness and alphabet knowledge are also associated with nonword reading. Nonword reading, or pseudoword decoding, involves reading nonsense words like “shap” or “tope” that follow the orthographic rules of a language. Nonword reading is a strong correlate of decoding skills for TD children (r=0.72, National Early Literacy Panel, 2008) and has also been found to be a weakness for children with a specific learning disorder with impairment in reading, also referred to as dyslexia (Ehri & McCormick, 1998; Rack et al., 1992). Thus, findings of strong nonword reading for children with ASD+HPL can serve as further evidence to support a phonological, or sublexical decoding approach to word reading for this group. In contrast, weak nonword reading might suggest a more whole word/orthographic approach to word reading. The findings are conflicted in preschoolers with ASD (without hyperlexia) where there is much debate in the literature surrounding the status of nonword reading skills. Some studies have found nonword reading on par with their TD peers (Smith Gabig, 2010; Knight et al., 2019) while others have shown that nonword reading is especially challenging (Westerveld et al., 2018). Consequently, in the debate over the status of nonword reading for preschoolers with ASD, TD preschoolers have served as a relative baseline comparison group, representing a typical, phonological approach to reading.

Other Behavioural Correlates of Literacy Skills

Previous studies have revealed a number of factors that may potentially influence emergent literacy skills and are thus taken into consideration in this literature review. For example, age of the participants may affect acquisition of phonological awareness, based on previous studies that show exposure to written instruction influences development in this area (Anthony & Francis, 2005). Therefore, school-aged and older children with ASD may have more advanced levels of emergent literacy based on previous exposure and instruction. ASD severity has been shown to predict phonemic awareness in preschoolers with ASD without hyperlexia (Nally et al., 2018) and is potentially linked to letter naming (Westerveld et al., 2017), thus may influence word reading. NVIQ has been shown to impact word reading for preschoolers with ASD without hyperlexia (Davidson et al., 2014; Westerveld et al., 2012). Similarly, VIQ, more specifically expressive language, was shown to be a strong predictor of reading ability in preschoolers with ASD (Davidson et al., 2014). Accordingly, the potential effect of these factors on emergent literacy in preschoolers with ASD and hyperlexia is taken into consideration.

Aims of this Review

Taken together, it is clear that there is much variation in conclusions drawn about competencies in phonological awareness, alphabet knowledge and nonword reading for preschoolers with ASD. Hence, we propose to review studies on preschoolers with ASD+HPL to examine if this is also the case for this population and identify variables that may account for any variation across studies. Therefore, the aims of this review are to 1) summarize the findings of studies of reading and early literacy skills with preschoolers with ASD+HPL which assess phonological awareness, alphabet knowledge, and nonword reading, 2) shed light on the potential reasons for discrepancies, if any, found across studies in word reading and early literacy skills and, 3) draw conclusions about the skills underlying word reading proficiency of preschool children with ASD+HPL (phonological/sublexical or an orthographic/lexical processes) to elucidate their reading trajectory and recommend intervention options. To do so, we propose a scoping review of studies that focus on emergent literacy skills, in particular phonological awareness, phonemic awareness, nonword reading and alphabet knowledge in the preschool population with ASD+HPL. We also examine the relationship between these variables and IQ, verbal IQ (VIQ), age and ASD severity.

Method

Search Strategy

According to Arksey and O'Malley (2005), a scoping review is operationalized as a way to summarize and disseminate research findings for the purpose of sharing this information with those who may not have the time to do so, but may benefit from this information. Scoping reviews are not as in-depth as systematic reviews and may be considered a first step in a process that warrants further investigation via a systematic review. To this end, a scoping review was chosen to examine the studies that assess the early literacy skills and nonword reading for preschoolers with ASD+HPL to provide an initial summary of emergent literacy skills for those working in the field of ASD+HPL research.

We used the PRISMA Flow diagram (Moher et al., 2009) as a procedural guideline (see Figure A1). Initial abstract searches were conducted on August 15th, 2019 carried out independently by two raters (the author and a colleague), with final searches conducted by the same two raters on June 10, 2020 in four electronic databases including: ERIC (EBSCO); PsycINFO (from 1806 to June 10, 2020); Medline© – Ovid Medline© Daily (including PubMed) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations Ovid Medliner Daily (1946 to June 10, 2020), and Scopus using the terms “Hyperlexia” OR “Hyperlexic”. These two search terms were used in two previous systematic analyses of hyperlexia (Ostrolenk et al., 2017; Zhang & Joshi, 2019).

The search was limited to articles published between January 1994 to June 2020. Studies prior to 1994 were excluded given changes in the Diagnostic and Statistical Manual of Mental Disorders (DSM) diagnostic criteria for ASD from the DSM-III (American Psychiatric Association, 1980) to DSM-IV (American Psychiatric Association, 1994). The DSM-III criteria for a diagnosis of ASD included “gross deficits in language development”, whereas the DSM-IV did not require a severe language deficit for diagnosis. Given that the purpose of the review is to explore language levels associated with ASD+HPL children, as approximately half of children with ASD have a language impairment, this approach was adopted to maintain an unbiased review with children with a severe language disorder, therefore, these earlier studies were omitted.

The search yielded a total of 227 articles with 30 from ERIC (Ebsco); 44 from Medline© (Ovid); 67 from PsycINFO 1987- (Ovid); and 86 from Scopus. All articles were imported directly into EndNote by both raters where duplicates were removed leaving 107 articles. Titles and abstracts of these 107 articles were reviewed resulting in removal of a further 59 articles independently by the two raters (see exclusionary criteria in the next section). The database included 48 full text articles assessed for final eligibility.

Exclusion Criteria

The 48 full text articles were assessed and excluded according to the following criteria: (a) Studies that did not have participants younger than 6 years. This decision centred on examining preschool children prior to any formal reading instruction. Given children with typical development can acquire phonological awareness in the absence of formal instruction, and that phonological awareness can be affected by formal instruction (Anthony & Francis, 2005), examination of children with ASD+HPL prior to any formal reading instruction offered the most insight into their true developmental skills associated with their advanced reading abilities; (b) Children with HPL who had a diagnosis other than ASD or a comorbid diagnosis with ASD. As HPL is comorbid with a number of neurodevelopmental disorders (see Zhang & Joshi, 2019) the intent was to focus only on HPL comorbid with ASD; and (c) Studies that were not peer-reviewed, unavailable in English or French, non-empirical, or provided insufficient assessment of reading skills or emergent literacy were also excluded. Following application of these exclusion criteria, and discussion between the two raters, 39 articles were removed, with a total of nine full text articles selected with 100% agreement between the two raters (see Figure A1). Thus, a total of nine articles were included for full review (see Table A2).

Data Extraction

Domain coding (Onwuegbuzie et al., 2016) was used to investigate, summarize, observe patterns and relationships among the following domains within each full text article (see Table A2): (a) Age and number of participants; (b) Level of acquisition of phonological awareness, alphabet knowledge (letter naming and phoneme-grapheme correspondence), and nonword reading, NVIQ, VIQ, ASD severity level; (c) Definitions, assessment measures and participants' native language; and (d) the relationship between participants' NVIQ, FSIQ, VIQ and ASD severity level and their associated proficiency in phonological and phonemic awareness, nonword reading skills and the alphabet knowledge.

Results

Phonological and Phonemic Awareness

Two studies of preschool participants with ASD+HPL (Cardoso-Martins & Ribeiro da Silva, 2010; Newman et al., 2007) examined phonological awareness skills. In the largest study (*N*=20) of children with ASD+HPL that included four preschoolers under the age of five and nine younger than 10 years old, Newman et al. (2007) assessed phonological awareness using the Sound Awareness subtest of the Woodcock Johnson Test of Achievement III (Woodcock, McGrew, & Mather, 2001). This subtest is composed of four parts: rhyming (identification and production), sound and syllable deletion (elision), sound and syllable substitution and reversal tasks (saying the sounds in a word backwards to create a new word, e.g., pit/tip). In terms of complexity, matching rhymes and syllable segmentation are early preschool tasks, while producing rhymes and sound deletion tasks are acquired in early to middle kindergarten respectively (Schuele & Boudreau, 2008). Newman et al. (2007) found the comparison group with TD (matched for reading ability, not for age, with the youngest participant being 6.6 years old) demonstrated stronger skills on these tasks as compared to the group with ASD-HPL, but not compared to the group with ASD+HPL. However, they reported that due to the composite nature of the Sound Awareness subtest (i.e., the various tasks of rhyming, sound deletion, sound substitution and reversal could not be scored separately); they were unable to evaluate the individual differences of each component of the subtest. They also determined that they could not accurately report on phonological awareness skills due to task complexity. In fact, only three of the youngest participants under the age of 10 years old in the group with ASD+HPL (*N*=20) were able to complete the Sound Awareness subtest.

Similarly, in Study 2 in Cardoso-Martins and Ribeiro da Silva (2010) two 3-year-old Portuguese preschoolers with ASD and reported hyperlexia were compared to 21 TD older preschoolers (*M*=6.14 years). Findings from their phonological awareness task with preschoolers with ASD+HPL were incomplete as the participants reportedly did not even attempt the task and all obtained scores of zero, while the older TD preschoolers performed at ceiling. Their tasks included a non-standardized, word initial sound identification task in Portuguese, whereby the children were asked to match a sound spoken by the examiner to a picture of a word beginning with that sound, from among a choice of three pictures. This initial sound identification task assesses phonemic awareness, and as such, is a more complex phonological awareness task (Schuele & Boudreau, 2008). Thus, it is possible that the phonemic awareness task was too difficult for the younger children with ASD+HPL. Furthermore, the performance of the older TD comparison group indicates it was an appropriate (and likely too easy) level for older children.

In summary, it is difficult to draw conclusions regarding the phonological awareness skills of the preschool ASD+HPL population since the skill was examined in only two studies as compared to an older TD comparison group, and task complexity is an issue with both phonemic and phonological awareness skills employed. In addition, both standardized and non-standardized measures were administered that complicate the task of cross-study comparison. Finally, orthographic depth of the two languages studied differs considerably, with English maintaining a deep orthography and Portuguese observing a shallower orthography. Languages considered to be shallow or surface orthographies, like Portuguese or Spanish, have a more regular one-to-one correspondence between orthography and pronunciation or phoneme-grapheme correspondences, e.g., mañana. In contrast, deep languages, like English tend to have more irregular orthography and pronunciations, e.g., yacht. Orthographic depth has been shown to affect the development of phonological awareness, with shallow orthographies accelerating the development of phonological awareness as compared to deeper orthographies (Francis & Francis, 2005). Thus, factors such as task complexity (phonemic versus phonological awareness tasks), standardized vs. unstandardized testing, participants' age and age of the TD comparison group, as well as orthographic depth of the participants' language may play a role in blurring our understanding of the acquisition status of phonological awareness for preschoolers with ASD+HPL.

Reading or Pseudoword Decoding

In this category, four studies were found which focused on nonword reading in preschoolers with ASD+HPL (Atkin & Lorch, 2006, Cardoso-Martins & Ribeiro da Silva, 2010; Lee & Hwang, 2015; Newman et al., 2007). Atkin and Lorch (2006) describe a single-case study where nonword reading was assessed using eight nonwords. The test consisted of both irregularly spelled words which violated English spelling conventions, (e.g., jvence), mixed with regularly spelled words with simple CV syllable structure, such as VC (ap), CV (dee) and CVCC (wip's) which included an apostrophe. Of the three nonwords the participant read both accurately and spontaneously (without instruction), all were regularly spelled nonwords (e.g., ap, dee, wip's). The participant did not accurately read any of the irregularly spelled nonwords correctly, e.g., pnir or jvence, nor the longer syllable structures, like wubfambit. Similarly, Lee and Hwang (2015) examined "real word-like nonwords" in Korean Hangul, where the nonwords only differed by one letter from a real word. They found that all participants read real words significantly more accurately than nonwords.

Other studies have examined regular nonwords compared to a TD comparison group (Cardoso-Martins & Ribeiro da Silva, 2010; Newman et al., 2007; Lee & Hwang, 2015) with similar outcomes. Cardoso-Martins and Ribeiro da Silva (2010) found children with ASD+HPL read a mean of 15 nonwords in Portuguese (out of 20) which was comparable to the children with TD who read a mean of 14 words. This finding was similar to that of Lee and Hwang (2015), as well as Newman et al. (2007) who found that the group with ASD+HPL was not significantly different from the TD group on regular nonword reading as assessed on the Woodcock Johnson Test of Achievement III, Word Attack subtest (Woodcock, McGrew, & Mather, 2001). However, there are several factors worth noting among these studies. Firstly, the WJ-ACH -III Word Attack subtest also includes grapheme-phoneme correspondence items prior to the nonword reading items. Thus, it is impossible to isolate the task of nonword reading using this subtest as a component of alphabet knowledge (grapheme-phoneme correspondence) is included in the task. Furthermore, only 4 of the 20 children with ASD+HPL in the Newman et al. (2007) study were preschoolers, and the results are not provided separately for this group.

Finally, the orthographic depth of the language in both the Cardoso-Martins and Ribeiro da Silva (2010) and Lee and Hwang (2015) studies contrasted with English. That is, both Portuguese and Hangul (Korean alphabet system) are shallow orthographies with regular and consistent spelling rules, and consistent grapheme-phoneme correspondence, as opposed to English, with its many spelling irregularities. Furthermore, in the case of the Cardoso-Martins and Ribeiro da Silva (2010) study, the authors did not list all the type of nonwords used.

Therefore, it is unclear whether the children in their study were reading nonwords with regular spelling patterns or patterns which violated the rules of Portuguese.

Taken together, these studies suggest that while preschoolers with ASD+HPL can read nonwords as well as their TD peers, there are several elements, including: validity of test items (testing only one construct); sample age (preschool versus school-age children); older TD comparison group; orthographic depth (shallow versus deep), and the type of nonwords used (regular, irregular, violating rules) that could influence outcomes for children with ASD+HPL for nonword reading when comparing across studies.

Alphabet Knowledge

Two studies examined components of alphabet knowledge in isolation (letter naming and phoneme-grapheme correspondence separately) in groups with ASD+HPL (Cardoso-Martins & Ribeiro da Silva, 2010; Lamonica et al., 2013). Both letter naming and phoneme-grapheme correspondence were assessed informally in Portuguese (Cardoso-Martins & Ribeiro da Silva, 2010). Participants were required to write all the letters of the alphabet; name them, write the letter associated with the letter name (when spoken by the examiner), and write the letter associated with the letter sound (when spoken by the examiner). Their results indicated that children with ASD+HPL could write all the letters of the alphabet and could write the letter when provided with the letter name, e.g., “bee” for ‘B’, with the same degree of accuracy as their TD peers (*M*=23 for both groups). However, the two groups differed significantly when they were required to write the letter associated with its sound, e.g., /b/ is for ‘B’. The group with ASD+HPL obtained a mean score of 8, whereas the group with TD obtained a mean of 21.52. Letter-name recognition was also assessed using informal assessments (Lamonica et al., 2013) with similar outcomes whereby all participants were able to recognize isolated letters of the alphabet when provided with the letter name with 100% accuracy.

To summarize, these findings suggest that the phoneme-grapheme correspondence skill, the hallmark component of alphabet knowledge, is potentially challenging for preschoolers with ASD+HPL in contrast to letter naming identification, which appears to be a strength. This is an important distinction as letter naming and phoneme-grapheme correspondence can be combined and assessed under the term alphabet knowledge. For example, the Alphabet subtest of the Test of Early Reading Ability – 3rd edition (TERA-3; Reid et al., 2001) assesses both phoneme-grapheme correspondence and letter naming together, as used by Davidson et al. (2014) in their study that examined this construct for preschoolers with ASD. Letter naming, as the name implies, is a confrontation naming task rather than a phonological awareness or metalinguistic task, as it does not involve the manipulation of sounds or syllables in spoken words, nor the transposing of letters to sounds as in grapheme-phoneme correspondence. Thus, combining these two different skills into one skillset may confound outcomes, especially since the findings in preschoolers with ASD (Knight et al., 2019; Nally et al., 2018) and ASD+HPL (Cardoso-Martins & Ribeiro da Silva, 2010) show that letter naming is a strength relative to phoneme-grapheme correspondence.

Age, ASD Severity, IQ and Language

Six studies (Atkin & Lorch, 2006; Cardoso-Martins & Ribeiro da Silva, 2010; Lamonica et al., 2013; Lee & Hwang, 2015; Lin, C-S., 2014; O'Connor et al., 1994) separately assessed preschoolers under the age of 6 years. All other studies (*n*=3) had a mix of preschoolers, with school-aged children (Grigorenko et al., 2002; Talero-Gutierrez, 2006) and adolescents

(Newman et al., 2007). Among all studies of preschoolers with ASD+HPL, none examined autism severity.

Six studies were found that included a measure of IQ (Atkin & Lorch, 2006; Grigorenko et al., 2002; Lee & Hwang, 2015; Newman et al., 2007; O'Connor & Hermelin, 1994; Talero-Gutierrez, 2006). Findings indicated no significant difference for IQ between groups with ASD+HPL and those with ASD-HPL (Grigorenko et al., 2002, Newman et al., 2007), which suggests that IQ is not a contributing factor to word reading. Similarly, Atkin & Lorch (2006) noted advanced word reading skills in a child with a mental age of 15 months that also suggests that word reading, and IQ are not related for children with ASD+HPL. Only one study reported that the mean word recognition age (*M*=7.1 years) of preschoolers with ASD+HPL (speakers of Korean Hangul) was not significantly lower than their FSIQ (Lee & Hwang, 2015) suggesting that there is a relationship between FSIQ and word reading. No studies of preschoolers with ASD+HPL explicitly examined correlations between IQ and word reading. In contrast, findings for preschoolers with ASD (without hyperlexia) suggest that NVIQ does contribute to word reading (Davidson et al., 2014; Westerveld et al., 2018). Of the four remaining studies: (a) two did not report VIQ nor NVIQ (Cardoso-Martins & Ribeiro da Silva, 2010; O'Connor & Hermelin, 1994), (b) one reported non-standardized measures (Lin, 2014), and (c) one did not include composite scores (Talero-Gutierrez, 2006).

Seven studies included measures of language (Atkin & Lorch, 2006; Grigorenko et al., 2002; Lamonica et al., 2013; Lee & Hwang, 2015; Lin, C-S., 2014; Newman et al., 2007; O'Connor et al., 1994). Comparisons of receptive and expressive language levels between groups with ASD+HPL to groups with ASD and TD revealed contradictory results, specifically for expressive vocabulary. That is, no significant differences were found for expressive vocabulary skills on the Expressive One Word Picture Vocabulary Test (EOWPVT; Martin & Brownell, 2011) for the group with ASD+HPL group (*M*=80.0) when compared with the group with ASD (*M*=89.7; Grigorenko et al., 2002). Yet, Newman et al. (2007) found that a group with ASD+HPL (*M*=110.55) performed better than the group with ASD (99.30), but comparable to the TD group (*M*=115.4) on the Picture Naming subtest of the WJ Test of Achievement III (Woodcock et al., 2001), a measure of expressive vocabulary. Thus, findings for expressive vocabulary of children with ASD+HPL are similar to those of TD children, but it is unclear whether expressive vocabulary is similar to or stronger than that of children with ASD without HPL.

Regarding receptive vocabulary, as measured on versions of the Peabody Picture Vocabulary Test PPVT (Dunn & Dunn, 1981). O'Connor and Hermelin (1994) reported findings in the average range for their two participants, which contrasted with findings of Cardoso-Martins and Ribeiro da Silva (2010) and Grigorenko et al. (2002) where lower mean scores were noted for the preschoolers with ASD+HPL as compared to TD peers and the group with ASD-HPL respectively. Although Lamonica et al. (2013) used a Portuguese version of the PPVT, they reported much heterogeneity within this skillset and only provided descriptors (e.g., lower low, higher low, lower average, etc.) rendering further interpretation difficult. Similar to the findings of O'Connor & Hermelin (1994), Newman et al. (2007) also found receptive language scores for the group with ASD+HPL to be on par with those of the TD group and the group with ASD-HPL using the Picture Vocabulary subtest of the Woodcock-Johnson Tests of Achievement – 3rd Edition (Woodcock et al., 2001). Therefore, like expressive vocabulary, there is much variation across studies in findings for receptive vocabulary, even when comparing across the same assessment measure.

Furthermore, there is some suggestion that receptive vocabulary levels are linked to phoneme-grapheme correspondence skills but not letter naming tasks. That is, preschool children with ASD+HPL with lower receptive vocabulary levels had no challenges with letter naming tasks but did struggle with alphabet knowledge (Lamonica et al., 2013) when compared to their TD peers (Cardoso-Martins & Ribeiro da Silva, 2010). No studies of ASD+HPL reviewed linked language abilities to word reading, in contrast with at least one study of preschoolers with ASD which demonstrated that VIQ, more specifically expressive language, was a strong predictor of reading ability (Davidson et al., 2014). Of the three remaining studies: (a) one study based their language assessment on their own assessment measure developed for the study (Lin, 2014), and (b) two studies provided a language age in place of standardized scores (Atkin & Lorch, 2006; Lee & Hwang, 2015).

In sum, it is unclear how ASD severity and NVIQ impact word reading in preschoolers with ASD+HPL, although NVIQ seems to contribute to word reading for children with ASD. Similarly, it is uncertain how expressive and receptive vocabulary skills of children with ASD+HPL compare to other preschoolers with ASD and TD preschoolers. However, there appears to be a link between a weakness in receptive vocabulary and lower levels of grapheme-phoneme correspondence skills for preschoolers with ASD+HPL that does not impact letter naming skills (although findings are limited).

Discussion and Recommendations

This scoping review focused on summarizing the findings across studies which examined the domains of phonological awareness, nonword reading and alphabet knowledge skills of children with ASD+HPL; and elucidating the factors responsible for potential discrepancies across studies. In addition, this scoping reviewed examined the impact of age, IQ and ASD severity on emergent literacy skills, and attempted to draw conclusions regarding the underlying skills supporting word reading proficiency in ASD+HPL. Finally, this scoping review aimed to clarify their reading trajectory as either similar to or different from the pathway used by their TD peers.

Reading Trajectory for Preschoolers with ASD+HPL

There are insufficient studies targeting emergent literacy skills for preschoolers with ASD+HPL and inconsistency of findings of the existing studies to make strong claims regarding the underlying basis of hyperlexia and whether it parallels or differs from the trajectory seen in their TD peers. The few studies that focus strictly on preschoolers with ASD+HPL typically have few participants. Grigorenko et al (2002) noted that studies of ASD+HPL are typically underpowered. Previous researchers have also noted that this group is particularly challenging to recruit (Newman et al., 2007). Our review reflects these assertions in the low number of studies and small sample sizes found for preschoolers with ASD+HPL.

Despite the limited studies, there are some emerging trends from the literature reviewed that are worth exploring in future studies. The alphabetic knowledge skill of phoneme-grapheme correspondence typically develops after phonological awareness skills in TD children, when they begin to connect spoken sounds to their written correlates (Scarborough & Brady, 2002). Indeed, phonemic awareness is a necessary prerequisite to the acquisition of phoneme-grapheme correspondences (Schuele & Boudreau, 2008). In fact, TD children who have not yet developed sufficient proficiency in phonemic awareness and alphabet knowledge are inefficient word decoders, relying instead on guessing based on the word initial sound and/or the syllabic structure of the word (Ehri, 2014). Yet, preschool children with ASD+HPL, who do not appear to have acquired the requisite alphabet knowledge task of phoneme-grapheme correspondence, still read words as well as older TD children and better than those with ASD (Grigorenko et al., 2002). These findings tend to imply that children with ASD+HPL are not relying on the same emergent literacy skillset as their TD peers. However, clearly more research in this domain is required to confirm these initial presumptions.

Phonological Awareness: Task Complexity or Lack of Acquisition?

It is difficult to draw conclusions concerning the acquisition status of phonological awareness for preschoolers with ASD+HPL. It has been proposed that the differences in complexity between tasks may be the source of variation seen in the groups with ASD (Westerveld et al., 2017) and this review suggests that is plausible. That is, developmental appropriateness of the phonological awareness tasks varied across studies and could have affected performance, as some studies targeted more complex phonemic awareness tasks. In a longitudinal study of TD preschoolers aged 3- to 4-years old, syllable and word-level rime awareness developed prior to phonemic awareness (Carroll et al., 2003). Thus, tasks that include phonemic awareness activities, focused on sound manipulation, may be too difficult for preschoolers and may not fully assess their actual abilities. Consequently, what might initially appear like a lack of phonological awareness skills, in fact, may be a result of assessing developmentally inappropriate phonemic awareness tasks. To this end, it is recommended that future studies match the age of the TD comparison group with the age of the group with ASD+HPL, rather than comparing to older TD children, or matching on reading ability, to fully contrast the emergent literacy skills of these two groups prior to formal reading instruction.

It has also been proposed that a lack of phonological awareness acquisition may be correlated with poor language acquisition such that children do not have adequate linguistic skills to understand the task instructions. However, based on the studies reviewed, there is no indication that receptive language skills are tied to phonological awareness, although this was not explicitly assessed in any study. Thus, previous findings which suggested that phonological awareness is impacted by a lack of comprehension of task instructions (Zhang & Joshi, 2019) cannot be supported by the findings of this review but remain to be determined. To test the hypothesis that phonological awareness skills may be affected by a lack of comprehension of task instructions or task complexity, tests of receptive phonological awareness which incorporate visuals, and have simple task instructions focused on early phonological awareness skills (rhyming and segmentation) may be more appropriate measures to employ. Receptive measures of phonological awareness have been found to be associated with improved performance and better comprehension of task instructions (Kenner et al., 2017). Thus, receptive phonological awareness measures may offer a way to tease apart the question of task complexity versus comprehension of task instructions, while offering a more developmentally appropriate means of assessment. In addition, examining correlations between receptive vocabulary or language acquisition and phonological awareness would also address this question.

Operationalizing Terms: Nonword Reading and Alphabet Knowledge

The need to operationalize the terms nonword reading and alphabet knowledge to improve reliability and validity for cross-study comparison was a primary outcome of this review. Regarding nonword reading, one issue centres on regular word reading versus nonword reading. For example, in at least one study nonword reading was operationalized as including both word decoding and nonword reading (Atkins & Lorch, 2006). Yet, studies of preschoolers with ASD+HPL and ASD demonstrate that although preschool children can read nonwords as well as older children with TD (Atkin & Lorch. 2006; Cardoso-Martins & Ribeiro da Silva, 2010; Lee & Hwang, 2015; Newman et al., 2007), regular words are easier to decode than nonwords (Newman et al., 2007; Westerveld et al. 2018). Thus, future studies should be aware that combining regular word reading and nonword reading may miss the opportunity for a more nuanced view of decoding abilities.

Furthermore, a number of different types of nonwords and words were used across studies. Differences included regularly spelled nonwords, irregularly spelled nonwords, nonwords with shorter syllable structure (CVC), nonwords with longer syllable structure (CCVC), and regularly spelled nonwords with apostrophes (Atkin & Lorch, 2006; Cardoso-Martins & Ribeiro da Silva, 2010). Indeed, nonwords used in at least one study (Atkin & Lorch, 2006) violated English spelling rules, thus potentially explaining the challenges participants experienced reading such words. Consequently, it is recommended that future assessment of nonword reading be conscious of these differences and either limit assessment to regular orthography that does not deviate from conventional spellings of the language or analyse these words separately, if included.

Standardized measures also contributed to inconsistent operationalization of the term by variably isolating the specific skillset under question. For example, the Word Attack subtest of the Woodcock Johnson Test of Achievement IV (Wechsler, 2012) includes phoneme-grapheme correspondence tasks with nonword reading. The implications of this are twofold. Firstly, there is some evidence to indicate that acquisition of these separate skills may be differentially acquired and therefore, differences become lost when averaged into one score. Secondly, total scores reported for nonword reading on this standardized measure may be under- or over-inflated dependent on how performance on one of these skillsets affects the other. Thus, future consideration of individual subtest items will permit greater specificity in assessment and facilitate cross study comparison.

Like nonword reading, consistent operationalization of the term Alphabet Knowledge across studies was also found to be a factor. Alphabet Knowledge has been regularly defined as both phoneme-grapheme correspondence skills alongside letter naming skills, as in the Alphabet subtest of the Test of Early Reading Ability – 3rd edition (TERA-3, Reid et al., 2001). Yet, this review suggests that these two abilities should be assessed separately, at least for preschoolers with ASD+HPL, given weaker phoneme-grapheme correspondence acquisition than letter naming. This discrepancy highlights the need to separately assess these tasks rather than combining them under one category of alphabet knowledge as per Scarborough and Brady (2005).

The Impact of Language

Orthographic Depth

Characteristics of diverse languages and orthographies across studies, more specifically, orthographic depth, may have contributed to the disparate findings for phonological acquisition for preschoolers with ASD+HPL. Although there is some debate in the literature as to the best characterisation or definition of orthographic depth (Schmalz et al., 2015), which is beyond the scope of this dissertation, generally languages have been characterized as shallow or deep. Of the studies reviewed, the two languages with the shallowest alphabetic or logographic orthographies are Portuguese (Defior et al., 2002) and Korean Hangul (Lee & Hwang, 2015) with regular phoneme-grapheme correspondences (or graphic-sound mapping in the case of Korean Hangul) that can potentially accelerate phonological awareness. Orthographic depth can affect the rate at which phonemic awareness skills are acquired and thus influence decoding of words in shallow orthographies versus deep orthographies (Anthony & Francis, 2005). In fact, the influence of orthographic depth on reading acquisition has been found across alphabetic, syllabic and logographic scripts (Ellis et al., 2004). Thus, it may be necessary to account for this when examining phonological awareness across languages and orthographies.

Like phonological awareness, nonword reading also appears to be impacted by uneven language characteristics across studies. Although the majority of studies on hyperlexia (75.68%) are conducted with English monolinguals (Zhang & Joshi, 2019), just over half of the studies in this review were conducted in English. The remaining studies reviewed included participants whose first language was Portuguese, Spanish, Hebrew and/or Korean – all considered shallow orthographies, in comparison to the deep, and more irregular English orthography. Therefore, the language of study and the corresponding impact of orthographic depth on nonword reading in preschoolers with ASD+HPL cannot be overlooked and should be considered in future studies.

The Influence of Receptive and Expressive Vocabulary on Emergent Literacy

There is some evidence from studies of TD children and those with ASD that suggest language skills, specifically receptive vocabulary, affect acquisition of phoneme-grapheme correspondence skills (Carroll et al., 2003; Lanter et al., 2012). Moving forward, future studies should consider the impact of receptive vocabulary when examining this variable. Finally, the discrepant findings for expressive and receptive vocabulary, as well as their relationship to phoneme-grapheme correspondence skills and phonological awareness, and the meaning of this relationship merits further investigation.

Additional Factors: ASD Severity, Age and IQ (NVIQ and FSIQ)

Outcomes from this review suggest that autism severity, age, IQ (at least NVIQ) may play a role in the divergent findings for phonological awareness acquisition across studies. Unfortunately, there were no studies examining the effect of autism severity on phonological awareness development. Studies of preschoolers with ASD have found severity predicted phonemic awareness (Nally et al., 2018) and is potentially linked to letter naming (Westerveld et al., 2017). Thus, future studies examining ASD severity in relation to emergent literacy skills in preschoolers with ASD+HPL are certainly warranted. Moreover, age range in a number of studies of ASD+HPL varied widely. For example, in the largest study (Newman et al., 2007), ages ranged from 3.0 years to 19.75 years. The inclusion of older children and adolescents has the potential to affect outcomes given the effect of exposure to written material on the development of phonological awareness (Anthony & Francis, 2005). Clearly, more studies focused solely on preschoolers and preschool-level phonological awareness tasks are required.

Finally, the variance across IQ and NVIQ levels for those with ASD+HPL in the presence of strong word reading skills, suggests IQ and word reading skills develop separately. However, as yet, no study has examined the correlation between IQ and word reading therefore, this remains to be examined explicitly. As there is some indication in young children with ASD that NVIQ may influence word reading (Davidson et al., 2014; Westerveld et al., 2012) it is suggested that the relationship between NVIQ and reading in preschoolers with ASD+HPL be examined more closely as well.

Clinical and Educational Implications

The importance of increasing our understanding of the early literacy skills of children with ASD+HPL is centred on early education. As children with ASD are included in regular classrooms (Vakil et al., 2009) it is important to understand whether teaching methods favour their academic development. The focus in kindergarten classrooms is on emergent literacy skills, like phonological awareness and alphabet knowledge, to develop word reading skills. If children with ASD+HPL are taking an alternate route, then teaching practices should reflect this to facilitate development. For example, an alternate route may consist of a more sight word or pattern-based approach to reading that might also inform other areas of learning and facilitate acquisition in associated domains like receptive and expressive language.

The benefits of a greater understanding of the mechanisms at play in their strength in early reading can also inform early interventions used by parents at home, as well as by clinicians and teachers at the preschool and kindergarten level. Further studies of preschoolers with ASD+HPL will deepen our understanding of the underpinnings of hyperlexia in ASD, to support and establish evidence-based practice. There is much to be learned from the 6%-20% of children with ASD who present with hyperlexia. Armed with this knowledge, we can better harness their strength in word reading, from the first moment it emerges as a special interest to when children enter school, to specify treatments that will best serve their needs.

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Appendix A

Figure A1

PRISMA 2009 Flow Diagram

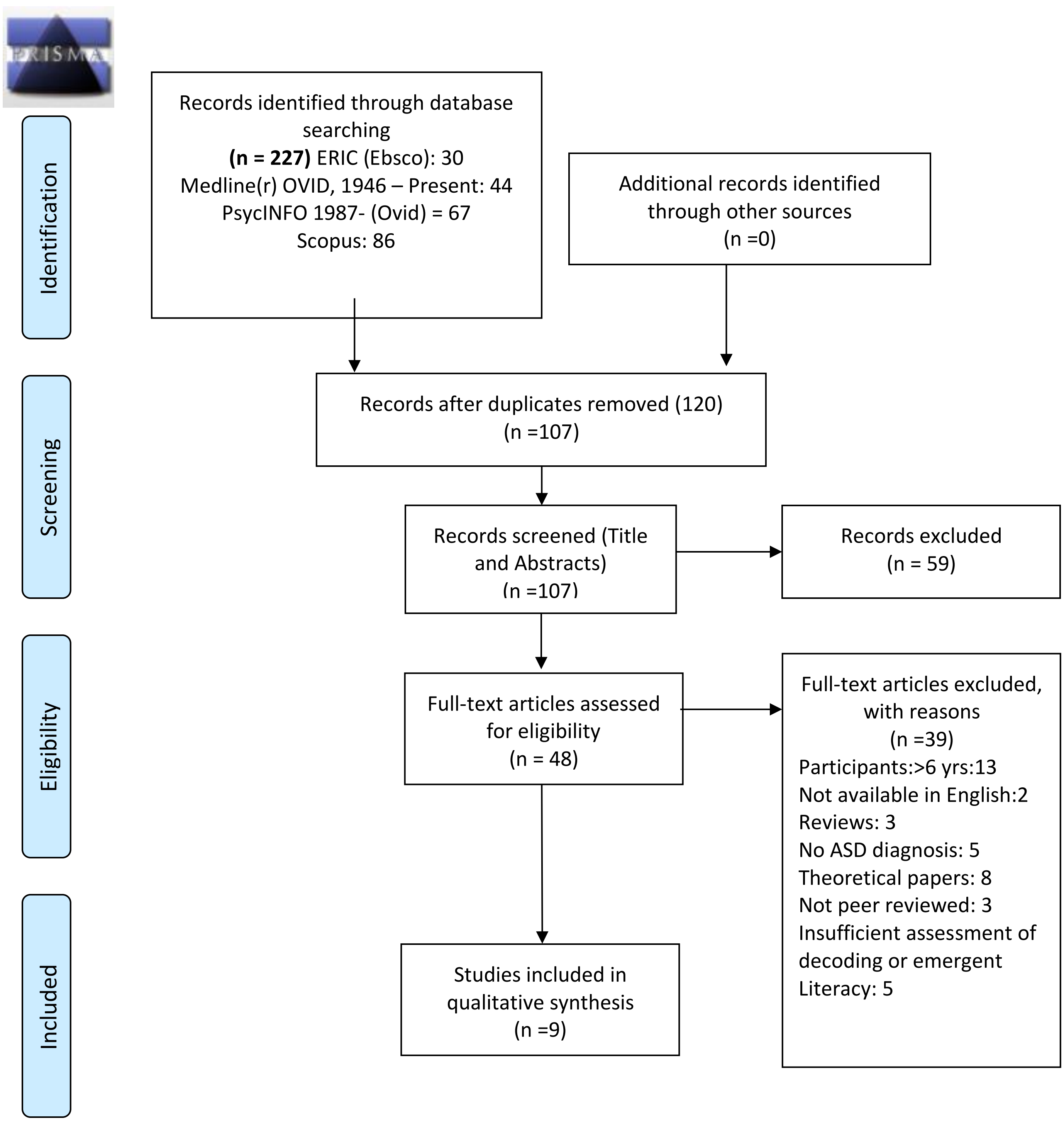

Table A2

Summary of studies included in the scoping review.

Author (Year)	**Participants Age and *N***	**NVIQ**	**VIQ**	**ASD severity**	**PA**	**Nonword Reading (NWR)**	**Word Reading (WR)**	**Alphabetic Knowledge (AK)**	**Results/ Author's Conclusions**
ASD+HPL									
Atkin et al. (2006)	**ASD+HPL** N=1 (male) CA: 4:3-4:7 yrs.	16 mths (at CA: 3:11) Griffiths Mental Developme nt Scale (GMDS, 1984)	GMDS 17 mths (at CA: 3;11). No spontaneous speech. L1=English	X	X	**NonStd test– 8 words (Healy, 1982) IRREG+REG** Had IRREG words that violated English spelling., e.g., pnir **3/8 REG NWD read correctly;** 5= close approximations (Aram, Rose & Horwitz (1984).	**REG+IRREG word pairs** (Aram et al., 1984), homophones (Ellis, 1993). Results: Read correctly: 12/18 REG words,10/18 IRREG words. 7/18 REG-IRREG-word pairs. 2 homophone pairs. Multisyll words were more difficult 6/17.	X	Learned IRREG pronunciations to pseudowords demonstrates advanced decoding skills. But spontaneous reading reveals challenges with IRREG NWR, and multisyllable words. There is linguistic processing of the text occurring given his paraphrasing when reading. Suggests an atypical route to language acquisition.
Cardoso-Martins et al. (2010) **Study 2**	**ASD+HPL** N=2 (males), CA:3.09 & 3:11yrs **TD,** N=21 (10 males) CA: 5.08-7.01 yrs. (*M*=6.14, SD=.52)	X	**TD>ASD+HPL** PPVT-III, ASD+HPL= 12.50 (11-14). TD=92.76 (16.64). Impaired VIQ in ASD+HPL L1=Portuguese	X	**TD>ASD+HPL,** *M*=0, TD, *M*=11.29 (1.06) Nonstd task: WI sound ID from a choice of 3 pictures. **ASD+HPL** did not attempt the tasks, hence 0 scores.	**ASD+HPL=TD** **NonStd**:20 Portuguese NWR ASD+HPL, *M*=15, TD, *M*=14.29 (3.55) Unclear if REG or IRREG (not all the words are listed)	**ASD+HPL=TD** Test of Achievement Stein (1994) Assessed single words (70 words) ASD+HPL=41.0, TD=41.43 No significant difference for all grps.	**ASD+HPL=TD on Letter Naming** No significant difference between grps. **TD>ASD+HPL on Letter-Sound Correspondence** ASD+HPL, *M*=8* (5-11), TD, *M*=21.52* (1.54).	Support a sound letter approach to reading based on phonological errors in reading despite findings of poor PA. Based on NWR and reading errors (phonological errors). They read by phonological recoding. No evidence of stronger PA in the HPL group. They query whether all the participants in Newman's (2007) were HPL's. They used Healy et al (1982) definition of HPL.
Grigorenko et al. (2002)	**ASD** N=79, CA: 2.67-12.50 yrs. (*M*=6.44, SD=2.35). **ASD+HPL** =12 Male=68 Severe DD **ASD-HPL,** N=67	**ASD+HPL =ASD-HPL Leiter** (HPL=70.50 and no HPL=81.79. K-ABC HPL=88.5noHPL=86.79	**ASD-HPL>ASD+HPL PPVT-R** HPL=56.7*, nonHPL=79.4* **ASD+HPL= ASD-HPL EOWPVT,** HPL=80.0, noHPL89.7 L1=English	X	X	X	**ASD+HPL>ASD-HPL** K-ABC. Sig dif btwn the HPL=111 and the nonHPL grp=94.91, *p*<.05	X	No significant difference btwn HPL and nonHPL for cognitive scores/IQ. Wide range of IQ in HPL. Sig diff on the PPVT btwn HPL and nonHPL. No sig diff btwn grps on EOWPVT. Higher prevalence HPL in boys

Lamonica et al. (2013)	**ASD+HPL** N=6 (males) CA:4;4-5;2 yrs.	X	**Heterogeneous ASD+HPL group** PPVT (Portuguese) Used descriptors: Lower AV; Lower Low; Higher and Higher AV. No scores L1=Portuguese	X	X	X	**Varied performance** School Performance Test. Descriptors related to performance in grade 1: BA, AV; AV. N=3(Higher), N=2(AV) and N=1(Lower)	**Strong Letter Naming** Assessed LN called Grapheme Recognition 100% recognition for all participants	All ASD+HPL LN = 100% accuracy. Heterogenous group in terms of VIQ and WR
Lee, 2015	**ASD+HPL** N=10, HPL Asperger's, n=2.CA:5;10 yrs. HPL dx based on discrepancy btwn word recognition and reading comp **TD**, N=10, CA:6;8 yrs.	K-WPPSI FSIQ, *M*=85.40	**Preschool receptive-Expressive Language Scale (PRES) –** Overall Language Age (LA). Word recognition> LA	X	X	**ASD+HPL=TD** 20 real word-like nonwords – all were 2 syllables and contained 4-6 letters. Each nonword had only 1 letter replaced from a familiar word. Higher mean error rates for nonwords (16%) than words (2%) – same for TD (21% and 0%)	**KEDI reading 1 subtest**. Recognize sight words. 20 high frequency words. More accurate at reading words than nonwords.	Letter naming on the KEDI reading subtest 1	No separate scores for LN provided.
Lin, C.-S (2014)	**ASD**, N=35, CA: 4-6 yrs. ASD+HPL n=15 (males)	X	**CALA** (Lin et al., 2013) Subtest 3 : VVC ; Subtest 4 : AVC	X	X	X	**ASD+HPL>ASD-HPL** 1SD>age-matched CALA subtest 1=Decoding; Subtest 2=Homographs (phrases) (decoding	X	They created their own test. Separated the ASD+HPL into different subgrps dependent on their performance on their test. NonStd measures are difficult to interpret.
Newman et al. (2007)	**ASD,** N=40 **ASD+HPL** N=20 (18= male) CA: 3-19.75, *M*=10.41 (4.65) Half (n=9) were <10 yrs. old. Only n= 4 were < or =5 yrs. **ASD-HPL** N=20 (18	**ASD+HPL =ASD-HPL** ASD+HPL WISC-III n=12; KABC n=3; Leiter-R n=1. IQ not available for n=4. *M*=99.4 (20.15).	**ASD+HPL= TD and ASD-HPL. TD>ASD-HPL** WJ ACH-III PV subtest. Sign grp diff F(2,55)=6.17 **	X	**ASD+HPL= TD and ASD-HPL. TD>ASD-HPL** WJ TA-III: SA subtest (rhyming, sound deletion, sound substitution, sound reversal); ***youngest participants had difficulty with the task**	**ASD+HPL>ASD-HPL** WJ ACH-III, WA subtest. Sign grp differences, *F*(2,54)=5.99**. But the actual difference was between the HPL and nonHPL grp, not btwn the HPL and TD or the nonHPL and TD where there were no sign differences.	**ASD+HPL=TD ASD+HPL>ASD-HPL** *p*<.03. WJ ACH-III, LWI subtest. WR SS and grp membership explained 73% of variance in reading comprehension (*F*(3,54)=49.13, *p*<.00 SS	This is included in the WA subtest of the WJ s but cannot be scored separately. Therefore, this was not assessed on its own.	1)PA skills were too difficult for the youngest members of the ASD+HPL grp and were not deemed valid measures 2) HPL grp has much "unevenness and variance" across their profile. This is especially true for single WR, comp and WA. 3) ASD-HPL group and TYP group are more similar. 5)The younger HPL grp performed better on WR comp and vocab than the older grp. The youngest HPL participants 5 and

	males), **TD**=18 (14 males), CA:9.99 yrs.	ASD-HPL, *M*=89.25; TYP (14 males) IQ tests not administere d.			; The TD > both the HPL and nonHPL grps *F*(2,36)=3.97** .	Strong NWR in the HPL grp.			under had the highest single WR scores. 6)Conclude based on their strong NWR, HPL grp relying on phonological decoding like TD readers.
O'Connor et al. (1994)	**ASD+HPL** N=2 (male) (CA: Time 1=5 and 8 yrs.; Time 2=9 and 12 yrs. Range: 5-7; 9-12 yrs.); **TD** (Time 2, N=2)	CA= 5 yrs. at Time 1, SS=138 on PM	PPVT=104 n=1, CA=5 at Time 1	X		X	Neale Reading Test	X	Focused on reading speeds. Reading comp comparable to cognitive development.
Talero-Gutierrez, C. (2006)	**ASD+HPL** N=2 (male) CA: 2-9;7 yrs. CA: 7-15 yrs.	Assessed @ CA: 9 on WISC-R, (ScS); Mazes:1; Coding: 12; OA:10, BD:6; PA:1; PC:1. No FSIQ	Important deficits in language expression and comprehensi on; Poor vocab No SS or ScS L1=Spanish	X	X	X	X	X	No composite scores for VIQ and NVIQ. Examine HPL in Spanish
<u>ASD</u>	Participants Age and N	NVIQ	VIQ	ASD	PA	NWR	WR	AK	Results/Authors Conclusions
Davidson, D.K et al (2014)	**ASD,** *N*=94 (n=82 males) Time 1: CA:*M*=30.84 mths (4.16) Time 2 CA:*M*=66.44 (4.96)	Mullen Early Scales of Learning: Nonverbal Ratio IQ Time 1, *M*=76.98	PLS-4 Time 1, AC *M*=60.83 (13.09). EC, *M*=72.73 (10.97)	ADI-R and ADOS or ADOS-T+Clinic al Judgeme nt	TERA-3 – Alphabet subtest included sound-letter correspondence, letter names, number of sounds and syllables and initial and final sounds. Scores not available for individual skills	X	*M* Reading Quotient on TERA-3 =88.64 (22.81), CA, *M*=5.5	TERA-3: Alphabet subtest SLC+LN *M*=11 (4.66). The alphabet subtest was significantly correlated with NVIQ**, SES**, social ability**, Rec** and Exp** language.	MESL *1)*NVIQ strong predictor of reading ability (AK)** 2) VIQ a strong predictor of reading abilities (Reading Quotient)** 3) Exp language not Rec language predicted overall reading ability* 4)(Time 1). The TERA Reading Quotient was negatively correlated (*r*=-22 with severity*
Dynia et al. (2017)	**ASD** *N*=108, n=35 (n=25 males) CA:52 mths (5.75) **TD**, n=73, (n=45 males)	**TD>ASD** ASD>4 yrs. (n=27), *M*=72.07 (21.48); TD, n=53,	**TD>ASD** CELF P2 ASD, n=32, *M*=68.31(19. 60); TD, n=70, *M*=95.07(6.9	X	**TD>ASD** TOPEL: EL, BL ASD, *M*=9.59(6.92); TD, *M*=15.41(5.68).P A significant	**WJ TA III: WA** Did not analyze mean difference btwn TD and ASD NonStd test: Assigned points to the # of pseudowords (32 possible total).	X	**ASD>TD** (PALS) – upper-case and lower-case ID). No SLC.	1)AK (*r*=0.52) and PA (*r*=0.41) significantly correlated with decoding (NWR)**. 2)ASD>AK than TD: ASD<PA than TD peers.

	CA : 52 mths, (7.25)	*M*=93.26 (14.08)	9).ASD Varied from average to severe		predictor (β=0.45) of decoding (NWR)*.	Referred to as decoding.			3)Oral language was sign correlated with all measures of emergent literacy** 4)Predictors of decoding in ASD are similar to TD.
Kimhi et al. (2018)	**ASD** N=5 (4 males), 5;9-7 ;4, *M*=82.20 mths; 1 child = 69 mths	X	PPVT-III *M*=75 (8.04) range = 63-95. L1=Hebrew	X	Nonstd test – SLC in Hebrew – Referred to as PA	X	X	LN in Hebrew	1) Found that "PA" (SLC) was poor at baseline, this could affect perceptions of PA for ASD (as it's not a PA task) 2) n=3 ASD had scores <70 on the PPVT-III also scored 0 on the SLC informal measure.
Knight et al. (2019)	**ASD** N=167 *M*=5.7 (80.7% male) Range 4-7,	Abbreviated version of the WPPSI-III. 3 subtest version *M*=88.45 (17.39) to estimate cog skills.	CASL: PC, BS; and SC subtests, *M*=81.97	ADOS convert to SVR Gotham Pickles and Lord's (2009) SS AIMSweb	**ASD<National Norms (1st grade)** PSF. Asked to provide all of the sounds in a given word. Did not do for PreK and K.	**ASD=National norms (1st grade) AIMSweb NWF**. nonsense words and asked to sound them out. Unclear if they are regular or irregular. WJ-ACH-III WA subtest correlated with NWF on the AIMS	WJ ACH-III, LWI did not do for the PreK or K.	**ASD>National Norms – LNF ASD<National Norms LSF (1stgrade)** AIMSweb: LNF (For children in pre-K, and K and grade 1); LSF for grade 1.	1)No sign diff btwn ASD severity grps for LNF in preK $t(62)$=.21, p=.84 2)LNF in PreK predicts reading comprehension. 3)SLC assessed for PA 4) Did not examine predictors of decoding.
Lanter et al. (2012)	**ASD** N=41, (n=33 males) CA: 4-8 yrs., *M*=69 mths	KBIT-2: Matrices *M*=79.0. TD = *M*=105.6; Mild-Mod, *M*=66.8, Severe, *M*=60.5	TELD-3. ASD grp into 3 grps: TD: *M*=96.3; Mild-Mod: *M*=66.8; Severe: *M*=43.7 *M*(grp)=64.6	X	X	X	Adapted ELP: EP subtest (identifying logographic images) and PC subtest (word-picture matching)	Adapted ELP Letter Identification subtest for LNI and LSC. No significant difference across subgroups for LNI p=.49 and LSC p=.12. Language was significantly associated with LNI* and LSC**	1)VIQ is related to both LNI and LSC. 2) Those with lower VIQ had lower levels of both LN and SLC. 3)Language level (severe impairment, mild-moderate impairment or typical) was found to be significantly correlated with LNI*and LSC** 4) Strong association btwn NVIQ and VIQ – query cognitive impairments.
Nally et al., 2018	**ASD** N=25 ASD alone; n=85 co-morbidity; (n=97 males) Group 1: CA: 3:10-5:10 Group 2: CA: 6.0-17.3	X	PLS-4 AC and EC EOWPVT-4 (*M*=82)	SCQ. Cut-off = 15	**Severely deficient** DIBELS – First Sound Fluency (FSF)=phonemic awareness, PSF = M=2.84phonological awareness, *M*=1.25	X	WIAT-II UK Ed. *M*=82.70	DIBELS: LN fluency, =9.54 (higher than FSF and PSF	1)Sign correlations btwn language and vocabulary, PSF, LNF and SVR 2)Sign correlation btwn word reading and LNF, phonemic awareness (FSF) and vocabulary. 3)Sign correlation btwn phonemic awareness and PSF, LNF and word reading 4)Sign neg correlation btwn SCQ and vocab 5) Age and SVR predicted word reading. 6) SVR predicted phonemic awareness.

Smith Gabig, C. (2010)	**ASD**, N=14, (12 males); CA range = 5;7-10 yrs; **TD**, N=10 (7 males); CA matched	**TD>ASD** DAS, ASD-HPL, *M*=96 TD, *M*=106. Sig diff btwn groups: *t*(22)=3.0** NVIQ not correlated with rec vocab or PA.	**TD>ASD** PPVT-III, Sign diff btwn the ASD and TD grp: *t*(22)=5.1	SCQ – Lifetime Cut-off =15	**TD>ASD** CTOPP – ELI and SBW subtests. Diff btwn groups, on ELI: *t*(22) =3.25** and BLW: *t*(22)=2.78*, ELI: ASD *M*=6, TD, *M*=10; For BLW: ASD *M*=8; TD *M*=11. ASD group scored below average compared to TD	**TD=ASD** WRMT-R: WA subtest. No sign diff btwn groups: *t*(22)=0.480 *p*>.05	**TD=ASD** WRMT-R; Word Identification subtest. No sig diff btwn grps:*t*(22)0.755, *p*>.05. Better decoding of real vs nonwords, *t*(13)=3.75**	X	1)WR and NWR were not related to PA. 2)ASD-HPL grp showed a bias towards sight word recognition over nonword reading suggesting the indirect nonlexical route to word reading is less developed. 3) ASD better able to read real words than nonwords but no difference for TD. 4)60% ASD struggled to read nonwords. 4)No correlation btwn PA and word recognition for the ASD grp. Concludes: PA is less well developed in the ASD grp.
Westerveld et al. (2017)	**ASD** N=57 (48 males).CA: *M*=57.63 mths (6.12). Range : 4 ;0-5 ;10 yrs.	MSEL – 2 subscales : VR & FM, *M*=79.11. Split grps along NVIQ with <70, N=21, and >70, N=36	PPVT-4, *M*=90.0 (16.3) VABS-II (Rec, Exp and Written). PPVT4 predicted code related ability (LN, LSK, PA, PWA and RAN)=0.093 , *t*=3.459**	SCQ-L (cutoff= 11 *M*=15.79 0 (5.753). r=0.255	PALS-PreK PA (word initial sound production) *M*=6.7 (total possible score =8). No sign diff btw groups for PA (p=0.071)	X	X	Adapted PALS-PreK (no discontinue rule applied): AK = LN, *M*=15.7 and LSK, *M*=8.6 Possible score of 26 on each test. Sig diff btwn groups on LSK** but not for LN (p=0.192	1)Relative strength in code related tasks (grouped together a number of these). 2)Did not state that LSK was weaker than LN though this was the case. 3)PA in the normal range (they suggest that initial sound ID could be easier for them). 4) No sig correlations btwn SCQ and any measures, though SCQ and LN approached significance, *p*=0.055
Westerveld et al. (2018)	**ASD** N=41 (35 males), Time 1 CA: *M*=57.6 mths (5.7). Time 2 CA: *M*=73.4	MSEL Time 1 measures as in 2017 study.	PPVT-4 (Time 1) CELF P2 (Time 2): STS, WS and EXPVOC subtests. AV readers do better on PPVT. *M*=99.4 compared to BA (*M*=80.1)**	SCQ-L Time 1 same as 2017 study. ASD sympto ms at Time 1 were not related to reading ability at Time 2.	PALS-PreK – Beginning sound awareness task AV readers (9.1) >BA Readers (4.2) on PA** No TD grp.	C&CT2 **Floor effects for nonword reading on the** Time 2 C&CT2: NWR. 19/40 children were unable to obtain a score on this measure (they selected reading as a measure instead given the strong correlation btwn REG and IRREG WR).	C&CT2: REG & IRR words. Single WR (Time 2), YARC: Passage Reading ability. Divided sample into AV readers (SS>85) and BA readers (SS<85). REG and IRR word reading strongly correlated** so used REG WR for single word ability. AV readers better on LSK, PA and NWR than "BA" readers	PALS-PreK LSK subtest AV readers (15.1) >BA Readers (3.7) on LSK** No TD grp	1)NVIQ and LSK accounted for 53.4% of the variance in REG WR at school age. 2) PPVT, Name writing and RAN accounted for 80.8% of the variance in reading grp (AV vs BA readers). 3)Language level impacts early literacy. 4) LSK was not a factor in their final model which they did not expect but was a predictor of single WR.

Note : AC=Auditory Comprehension; AK= Alphabet Knowledge; ASD=Autism Spectrum Disorder; ADOS=Autism Diagnostic Observation Scale; AVC=Auditory Vocabulary Comprehension; AV=Average; BA=Below average; BS: Basic Skills; CSS=Autism Diagnostic Observation Scale, Calibrated

Severity Scores; Bl=Blending; CA=Chronological Age; CALA=Computer Aided Language Assessment for Preschool Children with Autism; CASL=Comprehensive Assessment of Spoken Language; C&CT2=Castles and Coltheart Test 2; CELF P2=Clinical Evaluation of Language Fundamentals – 2nd Edition Preschool version; CTOPP=Comprehensive Test of Phonological Processing; DAS=Differential Ability Scales-Early Years or School Age; D=Deletion; ELI= Elision; ELP=Emergent Literacy Profile; EOWPVT=Expressive One Word Picture Vocabulary Test; EXPVOC=Expressive vocabulary; EP=Environmental Print; GMDS=Griffiths Mental Development Scale ; FM=Fine Motor; grp=group; GR=Grapheme Recognition: IRREG=Irregular; K-ABC = Kaufman Assessment Battery for Children; L1=first language; LC= Listening Comprehension; LN=Letter Naming; LNI=Letter-Name Identification; LNF=Letter Naming Fluency; LNI=Letter Name Identification; LSp=Letter Spelling; LS=Letter Sound; LSF=Letter Sound Fluency; LWI=Letter Word Identification; LSK=Letter Sound Knowledge; MSB=Multisyllabic; MSEL=Mullen Scales of Early Learning ; NRT= Neale Reading Test; NonStd=Non Standardized; NWR=Nonword Reading; NWF=Nonsense Word Fluency; OA: Object Assembly; PA=Phonological Awareness; PJ=Pragmatic Judgement; PC=Print Concept; PLS-4=Preschool Language Scale – 4th Edition; PM=Progressive Matrices; PPVT-R=Peabody Picture Vocabulary Test-Revised; PSF=Phoneme Segmentation Fluency; PV=Picture Vocabulary; REG=Regular; S=Segmentation; SA=Sound Awareness; SBW=Sound Blending Words; SC=Syntax Construction; ScS=Scaled Scores; SCQ=Social Communication Questionnaire; SI=Sound Identification; SLC=Sound-Letter Correspondences; SPT= School Performance Test; SRS=Social Responsiveness Scale; SS= Standard Score; STS=Sentence Structure; SVR=Severity Scores; ToA= Test of Achievement; TAP=Test of Academic Performance; TELD-3=Test of Early Language Development- Third Edition ; TOPEL=Test of Preschool Emergent Literacy; VR=Visual Reception; VVC=Visual Vocabulary Comprehension; WA=Word Attack; WJ TA-III=Woodcock Johnson Test of Achievement – 3rd Edition; WR=Word Reading; WRMT-R=Woodcock Reading Mastery Test-Revised; X=Not assessed or Not provided; YARC=York Assessment of Reading for Comprehension; * = significant at $p<.05$, **=significant at $p<.01$;()=Standard Deviation

Bridge Between Chapter 2 and Chapter 3

The primary goal of the literature review was to investigate the discrepancies and contraversies found across studies examining the early literacy profile of preschoolers with ASD+HPL, specific to phonological awareness, alphabet knowledge, and nonword reading. The purpose of clarifying the divergent findings was to identify factors which might account for cross-study discrepancy, establish a stronger empirical base for future research, and recommend teaching practices for facilitating reading development for children with ASD+HPL. The review also sought to examine the factors such as IQ, language, ASD severity and age that might influence acquisition. An additional goal was to clarify the early reading profile of preschoolers wth ASD+HPL as similar to or different from their peers with ASD-HPL and TD peers. Findings from the literature review contributed to advancing the research in the domain of emergent literacy and ASD+HPL by clarifying central issues requiring further research attention and illuminating the early reading profile of preschoolers with ASD+HPL. Further, findings from the literature review informed and directed a number of decisions made specific to the design and methodology of Manuscript 1 in Chapter 3 (referred to as Article 1 from this point forward).

For example, from the literature review it was clear that there was controversy surrounding whether preschoolers had adequate or weak phonological awareness as compared to their TD peers. More specifically, previous studies suggested that preschoolers may not understand the task demands of phonological awareness tests (Westerveld et al., 2017; Zhang & Joshi, 2019). In other words, task instructions or the task itself were possibly too complex or developmentally inappropriate for very young children leading to reduced performance, especially when compared to an older TD comparison group. Certainly, the results of the literature review demonstrated that some studies employed tests that assessed phonemic awareness that were likely too challenging for very young children. As a result, this was taken into consideration in Article 1 by including a chronological age-matched TD comparison group and by administering a visually based, receptive phonological awareness assessment measure (NEPSY-II; Korkman et al., 2007) that is recommended for children with learning differences, language and attentional delays and is standardized on children with autism. This standardized measure evaluates word segment recognition in 3–4-year-old children and adds phonological segmentation for those 5 years and older. These are phonological awareness tasks that are developmentally appropriate for preschoolers.

Findings from the literature review also suggested that alphabet knowledge skills were differentially acquired in preschool children with ASD+HPL. This implied that phoneme-grapheme correspondence skills, that support word reading in TD children (National Early Literacy Panel, 2008) were challenging for preschoolers with ASD+HPL, in contrast to letter naming skills, that appeared to be a strength. Based on this finding, Article 1 analysed phoneme-grapheme skills separately from letter naming.

Further to the examination of phoneme-grapheme correspondence, the literature review exposed issues with standardized testing that were taken into consideration in Article 1. The Word Attack subtest of the Woodcock Johnson Test of Achievement – Fourth edition (WJ-IV-ACH Word Attack subtest; Schrank et al., 2014) evaluates both nonword reading and phoneme-grapheme correspondence skills together. Considering the potential challenges that preschoolers with ASD+HPL have with phoneme-grapheme correspondence skills, it was decided to separate scores of phoneme-grapheme correspondence and nonword reading (using raw scores), rather than obtaining standard scores for the Word Attack subtest. This was done to avoid weaker scores for phoneme-grapheme correspondence negatively influencing the total nonword reading score (a reported strength).

Findings from the literature review also suggested that phoneme-grapheme correspondence skills might be linked to receptive vocabulary levels, at least for preschoolers with ASD with lower receptive vocabulary levels. Like phonological awareness, this would imply that task instructions were too difficult or potentially the tasks themselves were beyond their language ability. Thus, Article 1 also examined the correlations between receptive vocabulary and letter-sound correspondence skills in preschoolers with ASD+HPL.

Finally, Article 1 examined the correlations between ASD severity, age, IQ and early literacy skills and reading. Previous studies of young children with ASD (Davidson & Weismer, 2014; Nally et al., 2018) demonstrated a negative relationship between ASD severity and development of phonological awareness, such that increasing ASD severity was associated with weaker development of phonological awareness. Similarly, the literature review also served to examine the association between IQ and early reading skills. Building on these findings, Article 1 also examined the relationship between ASD severity and phonological awareness, as well as between IQ and early reading skills and preliteracy in preschoolers with ASD+HPL as outlined in Chapter 3.

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Chapter 3: Manuscript 1

This chapter is an exact reproduction of the published article (with the exception of tables 4, 5 and 6 appearing within the body of the text rather than in the appendices):

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Early Word Reading of Preschoolers with ASD, Both With and Without Hyperlexia, Compared to Typically Developing Preschoolers

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Abstract

A portion of children with Autism Spectrum Disorder (ASD) exhibit a strength in early word reading referred to as hyperlexia (HPL), yet it remains unclear what mechanisms underlie this strength. Typically developing children (TD) acquire phonological awareness, alphabet knowledge and language skills as precursors to word reading. We compared these skills across English-speaking preschoolers with ASD, both with and without hyperlexia, and TD preschoolers. Findings indicated that the group with both ASD and HPL (ASD+HPL) exhibited advanced word reading and letter naming skills as compared to the other two groups, but did not demonstrate commensurate phonological awareness, letter-sound correspondences, or language skills. Findings support an alternative, non-phonological approach to early word reading in preschoolers with ASD and hyperlexia.

Key words: autism, hyperlexia, phonological awareness, alphabet knowledge

Introduction

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by impaired social communication skills and repetitive patterns of behaviour (American Psychiatric Association, 2013). Among those diagnosed with ASD, a number of special abilities or strengths have been observed, including enhanced perceptual functioning (Mottron et al., 2006, 2009; Remington & Fairnie, 2017; Samson et al., 2012); enhanced pitch perception (Bonnel et al., 2003, 2010); musical memory (Heaton et al., 1999; Stanutz et al., 2014), and early word reading skills (Grigorenko et al., 2002; Needleman, 1982; Newman et al., 2007; O'Connor & Hermelin, 1994). This strength in early word reading was first referred to as "hyperlexia" by Silberberg and Silberberg (1967), characterized by a range of intellectual functioning from intellectual impairment to typical, along with word recognition skills higher than both reading comprehension and general verbal functioning.

Hyperlexia has been found primarily in an atypically developing population and comorbid with a number of neurodevelopmental disorders, including Attention Deficit Hyperactivity Disorder (ADHD), Tourette syndrome and specific language impairment (SLI) among others (Zhang & Joshi, 2019). Yet, ASD remains the most common neurodevelopmental disorder associated with hyperlexia (Ostrolenk et al., 2017). While the prevalence of hyperlexia is estimated to be 6%-20% among people with ASD, it is contingent upon the identification criteria (Ostrolenk et al., 2017). More stringent criteria, such as those put forth by Needleman (1982) that comprise five characteristics including having a developmental disorder alongside a very early and sudden manifestation of word reading, that is compulsive and ritualistic in nature and in advance of cognitive and other linguistic abilities, result in a lower prevalence rate as compared to definitions with fewer criteria (Ostrolenk et al., 2017). Currently, no consensual criteria are available to define hyperlexia across studies (Ostrolenk et al., 2017; Zhang & Joshi, 2019)

Despite the variability in criteria, previous studies have shown that children with ASD and hyperlexia demonstrate strong word reading in the presence of weak reading comprehension (Atkin & Lorch, 2006; Cardoso-Martins & Ribeiro da Silva, 2010; Grigorenko et al., 2002; Lamonica et al., 2013; Lin, 2014; Newman et al., 2007; O'Connor & Hermelin, 1994; Talero-Gutierrez, 2006). Reading comprehension is defined as the ability to derive meaning from written language passages (National Early Literacy Panel, 2008), while word reading is the ability to decode words using a phonological approach (i.e., sound-symbol/phoneme-grapheme relations) and/or using an orthographic approach (i.e., orthographic knowledge to verbalize sight words; National Early Literacy Panel, 2008).

Early Word Reading: Typical Development and ASD

For typical developing (TD) children, early word reading is predicated upon the initial development of phonological awareness skills, that is, the ability to manipulate sounds and syllables in spoken words (Ehri, 2009; Schuele & Boudreau, 2008). Phonological awareness develops from larger units (e.g., words and syllables) to smaller units (e.g., phonemes or sounds; Carroll et al., 2003). For example, segmenting the word "fireman" into two separate words (e.g., "fire" and "man") or the word "picnic" into two separate syllables, "pic" and "nic". Phonemic awareness, a more complex subskill of phonological awareness, requires attending to, and manipulating individual sounds in spoken words. Phonological awareness is classified as a metalinguistic ability that includes skills which reflect on, critically analyse and manipulate language (Scarborough & Brady, 2002). TD children as young as three years old have been shown to have developing metalinguistic abilities and competency in early receptive phonological awareness skills of segmenting and blending (Chaney, 1992; Kenner et al., 2017). With exposure to print, children acquire letter naming and letter-sound correspondence (grapheme-phoneme) skills, that is, the ability to assign sounds to the alphabet, collectively referred to as Alphabet Knowledge (National Early Literacy Panel, 2008). Alphabet knowledge enables decoding of regularly spelled words (Scarborough & Brady, 2002). For TD preschool and kindergarten children, alphabet knowledge is a strong correlate of word decoding skills (r=.50) while phonological awareness skills are a moderately strong correlate (r=.40) of word decoding (National Early Literacy Panel, 2008). In short, phonological awareness and alphabet knowledge are critical precursors to word decoding (i.e., reading single words) for TD children.

Yet, it remains unclear whether young children with ASD and hyperlexia are using the same precursors of phonological awareness and alphabet knowledge to read as do young TD children, or rather, are employing an alternate pathway uniquely their own. A model which has been used to account for superior word reading abilities for ASD is the Enhanced Perceptual Functioning theory (EPF; Mottron et al., 2006, 2009) in partnership with Veridical Mapping mechanisms (Mottron et al., 2013). The EPF model applied to word reading posits that pattern detection is heightened in ASD and that combining this strength in organized pattern or rule detection with an amplified interest and subsequently, overexposure to written material, rich in regularity and similarity, could lead to self-teaching or implicit learning of word reading in these very young children (Mottron et al., 2009). Veridical Mapping (VM) mechanisms are perceptual in nature and function to detect and pair similar patterns or codes to enhance learning (Mottron et al., 2013). In the case of hyperlexia, codes are the orthographic patterns of the language. For example, Consonant-Vowel-Consonant (CVC) is a common and basic syllable structure in English that, once acquired, can be extended to similar syllable structures to read numerous words, for example, cat -> hat, pan, pig, ten. This process is also referred to as orthographic recoding for TD children (Harris & Perfetti, 2017) and is based on Share's self-teaching hypothesis (Share, 1995, 1999), which facilitates learning of similar phonological codes. Thus, the EPF model, in combination with VM and orthographic recoding provide a model of early reading in ASD with hyperlexia via enhanced visual pattern perception applied to comparable orthographic codes, enabled by high exposure through an intense interest in letters and words. In support, results of parent questionnaires revealed that 42.50% of young children with ASD (mean age of 46.29 months) are more intensely interested in written material as compared to only 20.51% of TD children (Ostrolenk et al., 2018).

Reading, Phonological Awareness, Alphabet Knowledge and ASD, with and without Hyperlexia

There are a number of studies examining emergent literacy and/or early reading skills and young children with ASD without hyperlexia revealing mixed findings for phonological awareness acquisition. Some studies have found weaker phonological awareness skills for children with ASD (Kimhi et al., 2018; Knight et al., 2019; Nally et al., 2018), when compared with TD children (Dynia et al., 2017; Smith Gabig, 2010), with no relationship between phonological awareness and word reading (Smith Gabig, 2010). Others have found adequate phonological awareness (Kennedy, 2003) on par with typical development (Westerveld et al., 2017) and a correlation between phonemic awareness and word reading (Nally et al., 2018; Westerveld et al., 2018). It has been questioned whether these discrepant findings might be related to task complexity (Westerveld et al., 2017), as different tests measure developmentally different phonological awareness skills, or whether children possess adequate receptive language to understand the task demands of phonological awareness tests (Zhang & Joshi, 2019). In a recent meta-analysis examining hyperlexia across a range of age groups and neurodevelopmental conditions, Zhang and Joshi (2019) found that phonological awareness was related to listening comprehension, but not to decoding. This finding aligns with the observation that weak oral language skills have been documented for over half of children with ASD (Carlsson et al., 2013).

Few studies have examined emergent literacy and word reading for children with both ASD and hyperlexia. There are discrepant findings as to whether those with ASD with hyperlexia possess similar or impaired phonological awareness and alphabet knowledge as compared to their TD peers. Adequate phonological awareness skills for school-aged and adolescents with ASD and hyperlexia have been found (Kennedy, 2003; Newman et al., 2007; Saldaña et al., 2009). However, weak phonological awareness skills for very young children under age five have also been noted (Cardoso-Martins & Ribeiro da Silva, 2010; Newman et al., 2007)[1].

Regarding alphabet knowledge, findings across both groups of ASD with and without hyperlexia have generally indicated that letter naming is a strength, while letter-sound correspondence is a challenge (Cardoso-Martins & Ribeiro da Silva, 2010; Knight et al., 2019; Lamonica et al., 2013) or adequate but highly variable (Westerveld et al., 2017). Lanter et al. (2012) reported that letter naming and letter-sound correspondence skills for 4-5-year-old children with ASD without hyperlexia were positively related to language levels, with lower language levels associated with lower skills in both letter-sound correspondence and letter naming.

To our knowledge, no study to date has exclusively examined preschool children with ASD and hyperlexia (ASD+HPL) with sample sizes greater than two. Thus, we have yet to

[1] Newman et al., 2007 had four preschoolers aged 5 years and younger mixed with 16 school-aged and adolescents (*M*=10.41 years, range 3 – 19.75).

understand whether the word reading process of children with ASD+HPL is similar to that of TD children, that is, built upon preliteracy skills like alphabet knowledge and phonological awareness, or rather takes an alternative pathway. Examining preschoolers prior to receiving any formal reading instruction provides a view of word reading abilities in the absence of reading intervention. Critically, examining abilities at this early age would inform our knowledge of how young children with ASD process written material. Early examination is essential given phonological awareness for TD children is influenced by exposure to formal literacy instruction (Anthony & Francis, 2005).

The present study aims to 1) compare word reading and preliteracy skills of phonological awareness, alphabet knowledge (letter naming and letter-sound correspondence), and oral language skills among preschoolers with hyperlexia (ASD+HPL), without hyperlexia (ASD-HPL) and with typical development (TD); and 2) examine the correlations between these preliteracy skills within the three groups of preschoolers. We hypothesize that preschoolers with ASD+HPL and those with ASD-HPL will have weaker phonological awareness, alphabet knowledge and oral language skills than their TD peers. Furthermore, we expect phonological awareness, alphabet knowledge, and oral language skills will not significantly correlate with word reading for the groups with ASD+HPL and ASD-HPL, as they will for the TD group.

Method

Participants

Recruitment occurred through early childhood services for ASD; private psychology and speech-language pathology clinics; ASD and TD parenting Facebook groups; online ASD and TD parent group websites, and parenting magazines in Montreal and Ottawa, Canada. Participants were parents and their preschool children between the ages of 3 years to 5 years and

11 months, with an interest in letters, words, and/or an ability to read words, with no history of receiving formal reading instruction and who spoke English daily with at least one parent since birth. Fifteen participants were included in the group with ASD, of which 8 were included in the group with ASD+HPL and 7 in the group with ASD-HPL; 15 participants were included in the TD group. This study was approved by McGill University's Research Ethics Board.

ASD Symptoms and Inclusionary Criteria

Diagnosis of ASD was ascertained via documented comprehensive assessment reports conducted by registered clinical psychologists, paediatricians or psychiatrists (provided by parents or obtained with parental consent). In addition, the Autism Diagnostic Observation Schedule, 2nd edition (ADOS-2; Lord & Rutter, 2012); the Social Communication Questionnaire – Lifetime (SCQ-L; Rutter et al., 2003) and the Social Responsiveness Scale – Second Edition – Preschool Version (SRS-2P; Constantino, 2012) were administered. We used a cut-off *t*-score of 60 on the SRS-2P, and 11 on the SCQ-L, as suggested for young children (Moody et al., 2017). Scores above the cut-off fall in the ASD range and below in the TD range for both measures.

All participants with ASD (*n*=15) met criteria on the ADOS-2 by obtaining either a classification in the range of concern on the Toddler module, or at least a Mild-Moderate classification of autism spectrum on Modules 1, 2 or 3. One participant in the group with ASD+HPL had scores below the cut-off on both the SRS-2P and SCQ-L but met criteria for ASD on the ADOS-2. Two participants in the group with ASD+HPL did not meet criteria on either the SRS-2P or the SCQ-L but did meet criteria on the ADOS-2 and had an early word reading ability, thus were included in the group. All other participants in the group with ASD met criteria for ASD on the ADOS-2 and the SRS-2P or SQC-L (see Table 1).

Table 1

Age, FSIQ, VIQ and ASD Symptoms between groups (standard scores)

	ASD + HPL (n = 8)		ASD-HPL (n = 7)		All ASD Combined (N = 15)[2]		TD (N = 15)	
	M(SD)	Range	*M(SD)*	Range	*M(SD)*	Range	*M(SD)*	Range
Age (months)	53(9)	36–65	58(11)	41–70	55(10)	36–70	49(8)	38–61
WPPSI-IV FSIQ[a**c**]	69(8)	58–78	82(10)	67–94	75 (11)	58–94	87 (12)	68–112
WPPSI-IV VIQ[a**b*c**]	70(9)	58–85	88(13)	67–102	79(14)	58–102	95 (12)	71–111
WJ-IV ACH LWI[a**b**c**] Word Reading	121(14)	98–138	93(17)	72–110	95(14)	73–119	108(21)	72–138
ADOS-2 Comparison Score	7(2)	4–10	6(2)	4–9	7 (2)	4–10	–	–
SRS-2P[c**]	64(9)	54–78	74(7)	63–84	69 (9)	54–84	46 (6)	39–60
SCQ-L[c**]	13(6)	3–20	18(5)	12–24	16 (6)	3–24	8 (9)	1–29[1]

Statistically significant difference *$p < .05$, **$p < .01$ between: (a)ASD + HPL versus TD, (b)ASD + HPL versus ASD-HPL, (c)ASD versus TD
[1]Range of SCQ-L scores in the TD group without outliers is from 1–10
[2]All ASD Combined (n = 15) = ASD + HPL (n = 8) plus ASD-HPL (n = 7)

Parents of children in the TD group also completed the SRS-2P and SCQ-L. All participants but one (n=14) scored below the cut-off *t*-score of 60 on the SRS-2P indicating no risk of ASD. The one participant in the TD group that scored at the cut-off of 60 on the SRS-2P scored well below the cut-off on the SCQ-L. Two outliers were noted on the SCQ-L in the TD group with scores above the cut-off of 11; without these two outliers the range of scores for the SCQ-L would be 1-10. However, each of these participants scored well below the cut-off on the SRS-2P, and therefore, remained in the TD group.

All children in the group with ASD (N=15) had passed a hearing test documented in their diagnostic assessment report or reported by the parents as received under the newborn hearing screening program in Ottawa. Parents of six children in the TD group from Montreal reported their child never had a hearing test, however, all parents reported no concerns and only one parent reported a history of multiple ear infections. All parents of the remaining children in the TD group reported previous hearing tests indicating hearing within the normal range. No child presented with symptoms of hearing loss at the time of testing.

Hyperlexia Identification

The Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition (WPPSI-IV; Wechsler, 2012) was used to obtain a measure of full-scale IQ (FSIQ) using core subtests for each age group, and Verbal IQ using subtests of the Verbal Comprehension Index (VCI) reported as standard scores. The Letter-Word Identification (LWI) subtest of the Woodcock Johnson Test of Achievement – Fourth edition (WJ-IV-ACH; Schrank et al., 2014) was used to measure single word reading, reported in standard scores (see Table 1). For inclusion in the group with ASD and hyperlexia (ASD+HPL) five criteria based on Needleman (1982) were required: (a) "has an early manifestation" of single word reading, (p.475) before age 5 years, (b) a FSIQ below 78 (1.5 standard deviations below the normative mean), (c) a VIQ below 85 (1 standard deviation below the normative mean), (d) single word reading 2 standard deviations above VIQ and FSIQ (p. 479), and "minimal comprehension of written material" (p. 479). All participants, but one (*n*=7) in the group with ASD+HPL met all five criteria (see Table 1). One child in the group with ASD+HPL had reading comprehension on par with word reading and thus, did not meet the fifth criterion. The remaining children with a diagnosis of ASD were assigned to the group with ASD-HPL.

Study Exclusion

Initially, 37 children were assessed, with a total of seven children excluded from the study. One child from the TD group was excluded due to clinical signs of ASD and meeting criteria for hyperlexia. Six children were excluded from the group with ASD as they did not meet the ASD symptomology and/or hyperlexia identification for either ASD, ASD-HPL, or ASD+HPL, or could not be tested and were not exhibiting signs of hyperlexia.

Group Characteristics

Chi-square analyses revealed statistically significant gender differences between the groups with ASD and the TD group. Whereas the groups with ASD were largely composed of males (93%), the TD group was primarily female (73%). The high proportion of males in the groups with ASD reflects the sex discrepancy associated with ASD given that population estimates are of 4 males for 1 female with ASD (Baio et al., 2018) and is consistent with previous studies (Grigenko et al., 2002, Newman et al., 2007). There was no statistically significant difference on family income between the groups with ASD and TD (p=.33), nor between all three groups (p=.30).

In contrast, there were statistically significant differences between the groups with ASD and TD (n=30) on maternal education (p=.027) and between all three groups (p=.04), but not for paternal education (p=.47). There was no statistically significant difference in chronological age between ASD and TD groups (see Table 1). However, statistically significant differences were found on FSIQ, $F(1,28)=8.014$, $p=.008$, $\eta^2 = 0.22$, and VIQ, $F(1, 28)=11.90$, $p=.002$, $\eta^2 = .30$, with the TD group having higher scores on both FSIQ (M=87, SD=12) and VIQ (M=95, SD=12), than the group with ASD (FSIQ, M=75, SD=11; VIQ, M=79, SD=14) (see Table 1).

There were also statistically significant differences found on the SCQ-L, $F(1, 28)=8.74$, $p=.006$, $\eta^2 = .24$ and the SRS-P2, $F(1, 28)=62.27$, $p<.001$, $\eta^2 = .69$ with the group with ASD having higher scores on both the SCQ-L (M=16, SD=6) and SRS-2P (M=69, SD=9) than the TD group (SCQ-L, M=8, SD=9; SRS-2P, M=46, SD=6). An independent samples t-test revealed no statistically significant difference between the two subgroups with ASD on the SCQ-L or the ADOS-II comparison score, but there was a statistically significant difference on the SRS-P2 with the group with ASD-HPL having higher (more severe) scores $t(13)=2.39$, $p=.032$ than the group with ASD+HPL.

When comparing the three groups (see Table 1), there was a statistically significant group difference for FSIQ, $F(2,27)=7.43$, $p=.003$, and VIQ, $F(2, 27)=11.83$, $p<.001$. Post hoc analysis using Bonferroni correction revealed significantly higher mean FSIQ ($p=.002$) and VIQ ($p<.001$) for the TD group as compared to the group with ASD+HPL, but not the group with ASD-HPL. The group with ASD-HPL demonstrated significantly higher mean VIQ scores over the group with ASD+HPL ($p=.021$).

Materials and Procedure

In addition to the VIQ score obtained through the WPPSI-IV Verbal Comprehension Index (VCI; Wechsler, 2012), receptive vocabulary was measured using the Receptive Vocabulary subtest on the WPPSI-IV as part of the core subtests for children younger than four years old, and as a supplementary subtest for those older than four years. The Phonological Processing subtest of the NEPSY-II (Korkman et al., 2007) was used to provide a measure of phonological awareness. The NEPSY-II was chosen as it is a receptive measure of phonological awareness, assessing word segment recognition for 3-4-year-old children and adds phonological segmentation for those ages 5 years and older. The task of word segment recognition is a developmentally appropriate level of complexity for preschoolers as phonological awareness proceeds from larger units (words, syllables) to smaller units (phonemes) in TD preschoolers (Carroll et al., 2003). The NEPSY-II is recommended for children with learning differences, language and attention delays. In addition, standardization considered special group studies of children with autism (Brooks et al., 2009; Korkman et al., 2007).

The Letter-Word Identification (LWI) subtest of the WJ-IV-ACH (Schrank et al., 2014), the Word-Attack (WA) and Passage Comprehension (PC) subtests were also administered. The WA subtest provided a measure of letter-sound correspondence (using raw scores on items number 3-8) and pseudoword reading (using raw scores on item numbers 9-32). As only three participants (all in the group with ASD+HPL) were able to read pseudowords, this measure was not used in analyses. The LWI subtest provided a measure of letter naming (using raw scores on item numbers 1-10), and single word reading (using raw scores on items number 11 through 78). Three participants in the TD group and 1 participant in the group with ASD-HPL were excluded pairwise from the analysis of word reading as they obtained a ceiling (6 consecutive errors) within items 1-10 (letter naming) and therefore, never reached item #11 (the first item in word reading) on the LWI subtest. The WJ-IV ACH PC subtest is a measure of reading comprehension and lexical knowledge (Shrank et al., 2014) which begins with symbol representation, that is, matching black and white line drawings of objects to coloured images of the same object (items 1-4), and progresses to single word-, phrase-picture matching and sentence completion tasks. Because TD children, who were not yet reading, participated in these activities we chose to refer to this task as symbol and text representation, rather than reading comprehension.

The Oral and Written Language Scales – Second Edition (OWLS-II; Carrow-Woolfolk, 2011) Listening Comprehension (LC); Oral Expression (OE); and Reading Comprehension (RC) Scales were administered to measure receptive language; expressive language and reading comprehension (referred to as text-picture matching given most TD children were not reading yet), respectively. Versions B and C of the WJ-IV-ACH and versions A and B of the OWLS-II were randomly assigned to ensure counterbalance of the presentation order given that this study is part of a larger study where participants completed a different version of these tasks on three occasions. OWLS-II raw scores were used in analyses as standard scores are not available for children younger than 5 years of age on version B of the Listening Comprehension and Oral Expression Scales, as well as on versions A and B of the Reading Comprehension Scales.

Participants also completed two novel tasks as part of a custom-designed, tablet-based App. In-App tasks were comprised of an oral language-picture matching assessment task on the iPad as a measure of oral language (In-App Oral Language) and a Text-Picture Matching assessment task on the iPad (In-App Text-Picture Matching). For both tasks, participants chose a matching picture corresponding to a written word, phrase or sentence, from among a choice of four pictures. In the In-App Oral Language task the written word was accompanied by the corresponding auditory label. Raw scores were based on a total of 50 items for each assessment. All participants started at the first item and ended when a ceiling of three consecutive incorrect items was reached. The In-App assessment tasks were part of a larger study on the effects of a home-based, parent-supported reading intervention.

Data screening was performed prior to analysis. Outliers were assessed using boxplots. We retained all outliers as based on our small sample size; the outliers may represent actual variance rather than true outliers. We also noted deviations from the normal distribution, as assessed by a statistically significant Shapiro-Wilk test. We used the Kruskal-Wallis H Test with those variables containing outliers. Games-Howell non-parametric post hoc analysis was used when equal variances were not assumed. ANOVAs were used in analysis when all assumptions were met. Pearson's correlations were used to analyse the relationship between phonological awareness, alphabet knowledge, oral language skills and word reading in each of the three groups (ASD+HPL, ASD-HPL and TD).

Results

Language Variables

An ANOVA revealed statistically significant between-group differences (ASD+HPL, ASD-HPL and TD) on WPPSI-IV receptive vocabulary, $F(2,27)=4.29$, $p=.024$ and OWLS-II OE measuring expressive language $F(2,27)=5.85$, $p=.008$ (see Table 2, Figure 1).

Table 2

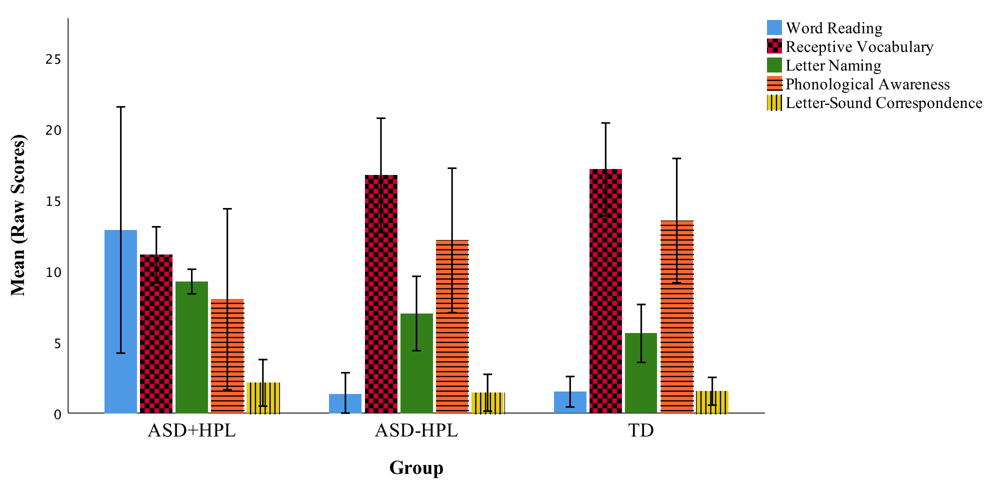
Group means on baseline language measures (raw scores)

	ASD + HPL ($n = 8$)		ASD-HPL ($n = 7$)		TD ($n = 15$)		All ASD Combined[1] ($n = 15$)	
	M (SD)	Range	*M(SD)*	Range	*M(SD)*	Range	*M(SD)*	Range
WPPSI-IV[a**b*] Receptive Vocabulary	11(2)	7–14	17(4)	10–23	17(6)	7–28	14(4)	7–23
OWLS-II Listening Comprehension (LC)	24(10)	10–45	36(18)	5–52	36(21)	16–87	29(15)	5–52
OWLS-II[a**b*c*] Oral Expression (OE)	9(5)	2–18	22(10)	8–38	24(12)	7–44	15(10)	2–38
In-App Oral Language	13(13)	0–36	20(12)	7–38	22(13)	2–44	16(13)	0–38

OWLS-II listening comprehension measures receptive language; *OWLS-II* oral expression measures expressive language
*Statistically significant difference $p < 0.05$, ** $p < 0.01$ between: (a) ASD + HPL versus TD, (b) ASD + HPL versus ASD-HPL, (c) ASD versus TD
[1]All ASD Combined ($n = 15$) = ASD + HPL ($n = 8$) plus ASD-HPL ($n = 7$)

Figure 1

Group performance on word reading, receptive vocabulary, letter naming, phonological awareness and letter-sound correspondence (raw scores) with error bars representing a 95% CI



Both receptive vocabulary and expressive language variables violated the assumption of homogeneity and therefore we used Games-Howell post hoc analysis, which showed statistically significant mean differences in scores between the TD group and the group with ASD+HPL on both receptive vocabulary (p=.007) and expressive language (p=.001), and between the groups with ASD+HPL and ASD-HPL for both receptive vocabulary (p=.034) and expressive language (p=.038), with higher mean scores in favour of both the TD group and the group with ASD-HPL over the group with ASD+HPL. There was no statistically significant difference between the TD group and the group with ASD-HPL on either variable. The Kruskal-Wallis H test revealed no statistically significant difference in the distribution of scores across groups for the OWLS-II LC measuring receptive language and the In-App Oral Language measure (see Table 2).

Phonological Awareness, Word reading, Letter Naming, Letter-sound correspondence, and Text-Picture Matching

An ANOVA revealed statistically significant differences across groups for single word reading on the WJ-ACH LWI subtest $F(2,23)=10.46$, $p<.001$, while the Kruskal-Wallis H test demonstrated statistically significant group differences in letter naming on the WJ-ACH LWI subtest $X^2(2)=6.76$, p=.034, text-picture matching on the OWLS-II-RC $X^2(3)=6.37$, p=.041, and the In-App Text-Picture Matching, $X^2(3)= 7.90$, p=.019. Post-hoc pairwise comparisons demonstrated that mean word reading scores for the group with ASD+HPL were significantly higher than both the TD group (p=.001) and the group with ASD-HPL (p=.004) (Table 3, Figure 1).

Table 3

Means, standard deviations, range and Ns for groups on baseline reading measures (raw scores)

	ASD + HPL (*n* = 8)		ASD-HPL (*n* = 7)		TD (*N* = 15)		All ASD Combined (*N* = 15)	
	M (SD)	Range	*M(SD)*	Range	*M(SD)*	Range	*M(SD)*	Range
NEPSY-II Phonological Processing	8(8)	0–18	12(5)	4–18	14(8)	0–22	10(7)	0–18
WJ-IV-ACH letter-Word Identification (LWI) Word Reading[a**b**] Items 11–78	13(10)	1–29	1(2)	0–4	2(2)	0–6	8(10)	0–29
WJ-IV-ACH LWI Letter Naming[a*] Items 1–10	9(1)	7–10	7(3)	2–10	6(4)	0–10	8(2)	2–10
WJ-IV-ACH Word Attack(WA) Items 3–8 Letter-Sound Correspondence	2(2)	0–6	1(1)	0–4	2(2)	0–5	2(2)	0–6
WJ-IV-ACH Passage Comprehension (PC) Symbol and Text Representation	6(4)	0–13	5(3)	3–11	5(1)	4–8	6(3)	0–13
OWLS—Reading Comprehension (RC) Text-Picture Matching[a*]	6(5)	0–15	2(3)	0–8	1(1)	0–4	4(4)	0–15
In-app text-picture matching[a*]	11(11)	1–29	3(2)	1–7	2(3)	0–12	7(9)	1–29

*Statistically significant difference = p < 0.05, **p < 0.01 between: (a) ASD + HPL versus TD (b) ASD + HPL versus ASD-HPL
NEPSY-II Phonological Processing measures Phonological Awareness; WJ-IV ACH = Woodcock Johnson Test of Achievement—Fourth Edition

The group with ASD+HPL also showed significantly higher mean scores on letter naming than the TD group (*p*=.028) but not compared to the group with ASD-HPL. Similarly, for text-picture matching in both the In-App (*p*=.015) and on the OWLS-RC (*p*=.036) the group with ASD+HPL demonstrated significantly higher scores as compared to the TD group, but not compared to the group with ASD-HPL. There were no statistically significant differences among groups in the distribution of scores for phonological awareness on the NEPSY-II, letter-sound correspondence on the WJ-IV-ACH-WA and symbol and text representation on the WJ-IV-ACH-PC.

Correlations between Language, Severity, FSIQ, Age and Reading Variables

The TD group showed statistically significant correlations between phonological awareness and all language measures (see Table 4, Figure 2), including WPSSI-IV receptive vocabulary (r=.747, p<.01), OWLS-II receptive language (r=.631, p<.05), expressive language (r=.723, p<.01), as well as letter-sound correspondence (r=.705, p<.01) and letter naming (r=.558, p<.05), but not with word reading (see Figure 3). Word reading was significantly correlated with letter naming (r=.652, p<.05) and text-picture matching on the OWLS-II (r=.774, p<.01) but not on the In-App text-picture matching.

Figure 2

Scatterplot of standard scores for receptive vocabulary (WPPSI-IV^{CDN}) and phonological awareness (NEPSY-II) for all three groups.

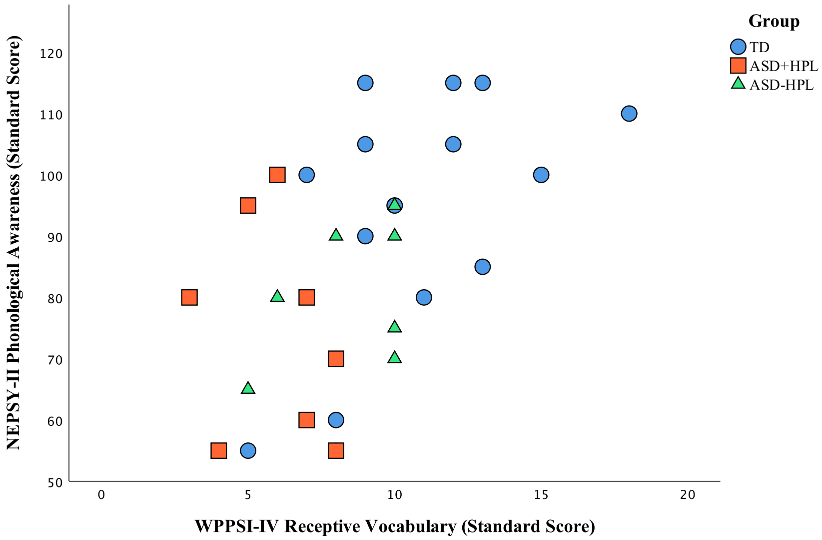
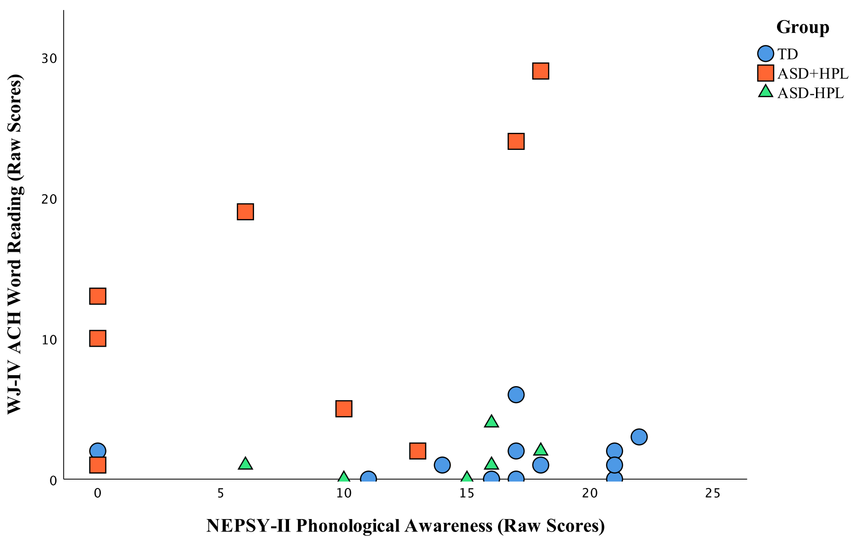


Figure 3

Scatterplot of raw scores on phonological awareness (NEPSY-II) and word reading (WJ-ACH-IV LWI) across all three groups



For the group with ASD+HPL, phonological awareness was significantly correlated with expressive language (*r*=.785, *p*<.05), letter-sound correspondence (*r*=.708, *p*<.05) and both the In-App (*r*=.724, *p*<.05) and OWLS-II-RC Text-picture matching (*r*=.728, *p*<.05), but not with word reading (see Table 4, Figure 3).

Table 4

Correlations for TD Group and Group with ASD+HPL (raw scores)

	TD								
	RV	PA	WR	RL	EL	LSC	LN	TTPM	IA-TPM
ASD+HPL									
Receptive vocabulary (RV)		.747**	.505	.893**	.935**	.883**	.749**	.284	.229
Phonological awareness (PA)	.454		.039	.631*	.723**	.705*	.558*	.168	.079
Word reading (WR)	.674	.536		.424	.425	.457	.652*	.774**	.279
Receptive language (RL)	.527	.633	.676		.856**	.839**	.671**	.300	.421
Expressive language (EL)	.274	.785*	.261	.619		.864**	.648**	.311	.421
Letter-sound correspondence (LSC)	.522	.708*	.691	.263	.398		.727**	.258	.239
Letter naming (LN)	−.190	.000	.296	−.074	.188	.335		.358	.423
Text-Picture Matching (TPM)	.561	.728*	.728*	.906**	.690	.363	.110		.423
In-App Text-Picture Matching (IA-TPM)	.496	.724*	.929**	.689	.406	.728*	.330	.785*	

Statistically significant *p < .05, **p < .01

Word reading was significantly correlated with both the In-App (*r*=.929, *p*<.01) and the OWLS-II Text-picture matching (*r*=.728, *p*<.05). Receptive language was significantly related to the OWLS-II Text-picture matching (*r*=.906, *p*<.01); however, neither receptive language nor receptive vocabulary were significantly correlated with phonological awareness (see Table 4, Figure 2). Letter-sound correspondence was significantly correlated with the In-App Text-picture matching task (*r*=.728, *p*<.05), but letter naming was not related to word reading nor any other variables. Thus, while the TD group had not yet developed word reading skills, their language was on par with phonological awareness and alphabet knowledge development, in contrast to the group with ASD+HPL, whose language, phonological awareness and alphabet knowledge were not commensurate with strong word reading skills.

Similar to the group with ASD+HPL, the group with ASD-HPL did not show a statistically significant correlation between phonological awareness and word reading nor any other variable (see Appendix Table 5, Figure 2). Unlike the group with ASD+HPL, the group with ASD-HPL did not demonstrate a statistically significant correlation between word reading and the In-App nor the OWLS-II text-picture matching tasks. Word reading was not significantly correlated with any other variable in this group.

Table 5

Correlations for groups with ASD-HPL and ASD+HPL (raw scores)

	ASD-HPL								
	RV	PA	WR	RL	EL	LSC	LN	TTPM	IA-TPM
ASD + HPL									
Receptive vocabulary (RV)		.672	.089	.714	.944**	.298	.285	−.238	−.101
Phonological awareness (PA)	.454		.409	.748	.670	.708	.741	−.395	−.335
Word reading (WR)	.674	.536		.299	.206	−.324	.023	−.285	−.029
Receptive language (RL	.527	.633	.676		.822*	.590	.550	−.141	−.149
Expressive language (EL)	.274	.785*	.261	.619		.349	.208	−.336	−.087
Letter-sound correspondence (LSC)	.522	.708*	.691	.263	.398		.717	.143	−.306
Letter naming (LN)	−.190	.000	.296	−.074	.188	.335		.227	−.053
Text-Picture Matching (TTPM)	.561	.728*	.728*	.906**	.690	.363	.110		.720
In-App Text-Picture Matching (IA-TTPM)	.496	.724*	.929**	.689	.406	.728*	.330	.785*	

Statistically significant $*p < .05$, $**p < .01$

We also examined age and ASD symptomology given the findings of Nally et al. (2018) and the differences we noted between groups with ASD on the SRS-P2. There was a statistically significant negative correlation (r=-.750, p<.05) between age and the SRS-P2 severity measure for the group with ASD+HPL, but not for the group with ASD-HPL (see Table 6). The group with ASD+HPL demonstrated decreasing (milder) severity with increasing age. However, neither age nor ASD symptomology was significantly correlated with either word decoding or phonological awareness. Finally, we found no statistically significant correlations between reading ability and FSIQ or phonological awareness and FSIQ for preschoolers with ASD+HPL, although there was a statistically significant correlation between phonological awareness and FSIQ for the group with TD group (*r*=.650, *p*<.001).

Table 6

Correlations for ASD severity, Age, Word Reading and Phonological Awareness for Groups with ASD+HPL and ASD-HPL (raw scores).

ASD+HPL	ASD-HPL					
	Age	ADOS-2	SRS-2P	SCQ-L	PA	WR
Age		.189	.050	.158	.525	-.004
ADOS-II Comparison Score	.368		-.560	-.309	.589	-.568
SRS-2P	-.750*	-.037		.825*	-.031	.578
SCQ-L	-.125	.174	.359		-.049	.117
NEPSY-II Phonological Awareness (PA)	.423	.215	-.573	-.300		.409
WJ-IV-ACH LWI Word Reading (WR)	.384	.348	-.622	-.162	.536	

Note: Significant **p*<.05, ***p*<.01

Discussion

The findings of the current study suggest that preschoolers with ASD+HPL have a strength in letter naming, but have not yet acquired metalinguistic skills, like phonological awareness and letter-sound correspondence to support reading words, despite their advanced word reading. Moreover, oral language skills for the group with ASD+HPL are not as well developed as their TD or ASD-HPL peers who have not yet begun to read. In fact, we found that oral language skills are not connected to word reading in the group with ASD+HPL, although expressive language (OWLS-II OE) and letter-sound correspondence (in agreement with Westerveld et al., 2018) are related to phonological awareness. These results contrast with those for older TD children where language and metalinguistic awareness are strong correlates of word decoding (National Early Literacy Panel, 2008). Our findings suggest that preschoolers with

ASD+HPL are likely not taking a phonological, language-based route to word reading as their TD peers, but rather an alternative pathway that relies less on phonological processing.

Phonological Awareness, Alphabet Knowledge, Oral Language and Word Reading

Our first hypothesis stated that preschoolers with ASD would have weaker phonological awareness, alphabet knowledge and oral language skills than their TD peers. This hypothesis was only confirmed for the group with ASD+HPL for oral language skills which were significantly weaker than their TD peers. Phonological awareness, and letter-sound correspondence were not significantly different from their TD peers, while letter naming was, in fact, significantly stronger. Regarding the group with ASD-HPL, unexpectedly, we found their profile appeared to be similar to that of the TD group across VIQ, receptive vocabulary, word reading and pre-literacy skills. Similarities across these two groups replicates findings by Newman et al. (2007). The Newman study found that Reading Comprehension scores distinguished the TD group from the two groups with ASD, which was not the case in our study. Although the Newman study did not examine FSIQ for their TD group, we noted that mean FSIQ for our TD group (M=87) was relatively low. Despite statistically significant differences in full scale (ASD+HPL vs. TD) and verbal IQ (ASD+HPL vs TD; ASD+HPL vs ASD-HPL), 40% of TD participants (6/15) demonstrated a FSIQ of less than 85, with three participants in the borderline range (70-79) and one below 69 in the extremely low range according to the WPPSI-IV^{CDN} descriptive classifications. We intentionally did not exclude children based on FSIQ given the wide range of FSIQ found in preschoolers with ASD+HPL (Grigorenko et al., 2002). However, participants in the TD group with borderline intellectual functioning may have contributed to overall lower mean scores for this group across the outcome variables, negatively skewing the distribution, and accounting for the similarity in profiles between the group with ASD-HPL and the TD group.

Our second hypothesis of no relationship between phonological awareness, alphabet knowledge, oral language skills and word reading within the groups with ASD was supported by our results. Previous studies have also found no accompanying relationship between these variables (Smtih Gabig, 2010) or reported they did not predict word reading in children aged six years and older (Nally et al., 2018). Furthermore, the group with ASD+HPL demonstrated the weakest mean scores on phonological awareness. Other studies have also found weak phonological awareness skills in young children with ASD (Cardoso-Martins et al., 2010; Dynia et al., 2017; Kimhi et al., 2018; Nally et al., 2018; Smith Gabig, 2010), as well as Newman et al. (2007) for the very youngest participants in their group with ASD+HPL (see Footnote 1).

Although all three groups demonstrated a similar profile for letter-sound correspondence within alphabet knowledge, the discrepancy between letter naming and letter-sound correspondence, in favour of stronger letter naming for the group with ASD+HPL, was particularly unusual given their advanced word reading. That is, there is evidence that letter-sound correspondence along with phonemic awareness causally influence early reading skills for young TD children (Hulme et al., 2012), and yet, preschoolers with ASD+HPL in our study had weak letter-sound correspondence in the presence of strong word reading. This serves as further evidence of an alternative approach to reading in preschoolers with ASD+HPL. Moreover, it also suggests that alphabet knowledge skills should be treated distinctly as letter-sound correspondence and letter naming, rather than combined, at least for the children with ASD+HPL.

Our finding of strong letter naming in the group with ASD+HPL has been widely reported in the literature (Cardoso-Martins & da Silva, 2010; Knight et al, 2019; Lamonica et al., 2013; Westerveld et al., 2017). However, unlike phonological awareness and letter-sound correspondence, letter naming is not a metalinguistic task requiring analysis of language, but rather a confrontation naming task (Scarborough & Brady, 2002) with a one-to-one correspondence between the name and the object. In contrast, phonological awareness and letter-sound correspondence tasks require linguistic analysis or metalinguistic awareness (Schuele & Boudreau, 2008). Our findings revealed no statistically significant difference among the groups in phonological awareness or letter-sound correspondence despite advanced word reading in the group with ASD+HPL, suggesting a potential lack of underlying metalinguistic development contributing to word reading in this group. These findings support an alternative approach to word reading and are consistent with hyperlexia being found in atypically developing populations (Zhang & Joshi, 2019).

Accordingly, the Enhanced Perceptual Functioning Model (Mottron et al., 2006, 2009), VM and orthographic recoding fit with our findings. Collectively they offer an explanation for the acquisition of word reading in the absence of any formal instruction, language correlates, phonological awareness or letter-sound correspondence skills in preschoolers with ASD+HPL. The strength in letter naming may reflect a more regular, pattern-based approach to reading based on high predictability and facilitated by regularity and similarity, rather than irregularity in the orthography. In contrast, the challenges noted in letter-sound correspondence and phonological awareness may be considered equivalent to an absence of the use of decoding via a phonological or sound-based approach. Hence, word reading may be seen more akin to a rote naming task founded on whole word orthography acquired by perceiving similarities across comparable syllable structures, rather than a true word decoding task focused on manipulating the individual sounds in words.

Phonological awareness: Understanding Task Instructions or a Lack of Acquisition?

Although we found receptive language (OWLS II-LC), phonological awareness and word reading skills were significantly associated with text-picture matching on both the In-App and OWLS-II Reading Comprehension subtest, we found no statistically significant correlation between phonological awareness and receptive language or receptive vocabulary in the groups with ASD which replicated Nally et al. (2018). In contrast we found phonological awareness was related to expressive language in the group with ASD+HPL replicating Davidson and Weismer (2014). Phonological awareness was also correlated with letter-sound correspondence in the group with ASD+HPL, and both these metalinguistic skills demonstrated weaker development than would be expected based on reading levels. That is, the group with ASD+HPL performed one standard deviation below the test's normative average on the NEPSY-II, a test recommended for children with language delays and learning differences, that incorporates developmentally appropriate phonological awareness tasks and considered children with autism in standardization. In contrast, our findings for the TD group are consistent with those found in the literature for TD preschoolers which demonstrate that receptive vocabulary is a significant correlate of early phonological awareness (Carroll et al., 2003). In addition, the TD group was the only group whose scores were within the normative average on the NEPSY-II.

Together, these findings suggest that performance on the phonological awareness task for the groups with ASD was not related to poor understanding of task instructions, but rather less well-developed phonological awareness skills of segmentation and letter-sound correspondence. Our findings are in contrast to the meta-analytic findings of Zhang and Joshi (2019) in which phonological awareness was found to be related to receptive language skills. However, their meta-analysis included hyperlexia comorbid with a number of other neurodevelopmental conditions, and included an older sample, which may have affected outcomes.

Additional Factors related to Word Reading: Age, ASD Severity and FSIQ

Unlike previous studies of preschoolers with ASD (Davidson & Weismer, 2014; Nally et al., 2018), we found that neither age nor ASD severity on the ADOS-II, SCQ-L or the SRS-P2 was significantly related to either word reading or phonological awareness (see Table 6) for both groups with ASD. Despite the group with ASD+HPL showing milder symptoms with increasing age on the SRS-P2, their reading did not increase correspondingly with age or less severe symptomology. In addition, FSIQ did not differ significantly between the groups with ASD+HPL and ASD-HPL in agreement with previous studies (Grigorenko et al., 2002; Newman et al., 2007). However, the mean FSIQ of 69 in our population of preschoolers with ASD+HPL was lower than groups with ASD+HPL in previous studies (Grigorenko et al., 2002; Newman et al., 2007) and lower than our group with ASD-HPL. Furthermore, the range of FSIQ from 58 to 78 in our group with ASD+HPL was narrower and lower than has been found in other studies (Grigorenko et al., 2002; Newman et al., 2007) and reported by Silberberg and Silberberg (1967) who noted a wide range in FSIQ from very low to average. Our findings are more in line with those found in Atkins and Lorch (2006) who reported a mental age of 1.5 years in a 4-year-old boy and Talero-Gutierrez (2006) whose patients could not complete standardized cognitive testing to obtain a FSIQ.

It has been proposed that lower IQ found for those with ASD may be associated with an elevated need for pattern-based interpretation as per the hyper-systemizing theory linked with IQ put forth by Baron-Cohen (2006), which would also align with the EPF model. In support, previous studies (Atkin & Lorch, 2006; Newman et al., 2007) have recounted how participants with ASD+HPL initially needed to “correct” the pseudowords by providing a regular pronunciation or an orthographically similar real word (e.g., got/gat) in place of the pseudowords they were asked to read. Similarly, in our study, only 3/8 participants with ASD+HPL were able to read pseudowords. Thus, preschoolers with ASD+HPL may be applying an early strategy of pattern detection to the orthography to compensate for challenges in other areas like cognition, language, phonological awareness and letter-sound correspondence.

Despite our findings of an alternative approach to word reading in preschoolers with ASD+HPL, there were several limitations to this study. Although we obtained a sample in our group with ASD+HPL greater than two, which was to our knowledge the largest sample to date for a study on ASD+HPL with preschool-age participants (Atkins et al., 2006; Cardoso-Martins & Ribeiro da Silva, 2010) our sample size remains small and thus limits our ability to generalize beyond our study sample. This speaks to the challenges that researchers face when studying this population as reported by Newman et al. (2007), especially when studying children under 5 years of age who may not yet have received a diagnosis of ASD. Indeed, only 42% of children with ASD have been fully evaluated by age 36 months (Baio et al., 2018) and at least one previous study reported a mean age of hyperlexia diagnosis of 5.9 years of age (Grigorenko et al., 2002). Therefore, it is not surprising that studies of ASD+HPL have been found to be typically underpowered (Grigorenko et al., 2002).

We also experienced limitations in the standardized measures used for the preschool population (Westerveld et al., 2017). For example, the WJ-IV ACH Word Attack subtest measures both letter-sound correspondence and pseudoword reading skills. While our preference was to use standardized tests to measure variables, there were only six items associated with letter-sound correspondence on the subtest. We chose to combine the two items together (pseudoword reading and letter-sound correspondence) to limit the number of tests administered to preschoolers. As only a few participants in the group with ASD+HPL were able to read pseudowords, we were unable to fully analyse the type (regular or irregular) of pseudowords read successfully. Similarly, the WJ-IV ACH Letter Word Identification subtest combined both letter naming with word reading. Given the findings of alphabet knowledge skill and preschoolers with ASD+HPL, future studies should consider examining letter-sound correspondence separately from letter naming in relation to regular and irregular word and pseudoword reading as a means to further elucidate the underlying mechanism supporting word reading in this population. Finally, the OWLS-II scales provide norms for three-year-old children in only one version (A) of their test for language, and not for reading comprehension, which limited our analysis to raw scores. However, the OWLS-II scales are picture-based and were engaging for the children with autism, therefore, despite this limitation they were an appropriate measure for our preschool population

Regardless of these limitations, our findings have implications for learning in the early years when preschoolers with ASD are at home, in preschool or in early kindergarten environments. As noted in Newman et al. (2007), parents report that school programs do not support the reading abilities of their child with HPL. Preschool and kindergarten classrooms are focused on phonological awareness and alphabet knowledge activities that foster early reading skills and represent best practices in early literacy development for TD children (National Literacy Panel, 2008). Yet, our findings suggest that these best practices do not fit the unique approach to reading of young children with ASD. Kindergarten teachers aim to teach reading skills to TD children, while preschoolers with ASD+HPL already demonstrate mastery of this task. This puts children with ASD+HPL at risk of losing their interest in reading, which may be related to findings of mean word reading decreasing with age as compared to TD peers (Newman et al., 2007). In addition, parents in our study reported that their child is frequently tasked with reading to other children in the classroom. While this is well-intentioned, their strength in word reading could be better applied to tasks which support associated challenges in their oral language development and reading comprehension, in place of more practice in word reading. This study represents the first steps towards launching evidence-based practice guidelines and appropriate interventions through a greater understanding of the mechanisms underlying strengths in word reading in preschoolers with ASD+HPL and ASD-HPL.

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Bridge between Manuscripts – Chapter 3 and Chapter 4

In Chapter 3, the main question addressed by Article 1 was, " Do preschoolers with ASD+HPL learn to read in the same way as TD preschoolers?". To this end, the underlying mechanisms that support word reading of preschoolers with ASD and hyperlexia were examined. From the literature review in Chapter 2, findings indicated that there were few studies exclusively focused on preschoolers and those that did generally had small samples of one or two participants. In addition, factors were identified that contributed to diverse findings including: task complexity, age of the participants, orthographic depth of the language, types of nonwords used, and varying definitions of constructs like alphabet knowledge. Therefore, Article 1 in Chapter 3 addressed a number of these issues in an effort to understand the nature of emergent literacy in preschoolers with ASD and hyperlexia.

Comparing preschoolers with ASD and hyperlexia to preschoolers with ASD without hyperlexia and age-matched, typically developing (TD) preschoolers, findings indicated that while preschoolers with ASD and hyperlexia have a strength in letter naming, they lack a strong base in language, phonological awareness and grapheme-phoneme correspondence skills that TD children use to decode words on a sound-by-sound basis. This finding suggested that preschoolers are not using a phonological route to word reading but are potentially relying on a pattern-based approach as per the hyper-systemizing theory, the Enhance Perceptual Functioning theory (Mottron et al., 2006, 2009) and Veridical mapping (Mottron et al., 2013). Their intense interest in print is potentially relevant to their conceivable strength in pattern analysis, in this case, directed at the orthography of a language, allowing them to make associations between similar orthographic patterns and read similar words. This deeper understanding of their early word reading skill led to recommendations for early reading instruction tailored to young children with ASD+HPL who are likely integrated into classrooms where TD children are receiving reading instruction focused on phonological awareness and phoneme-grapheme correspondence skills.

Subsequent to these findings, the focus shifted to the main challenge associated with hyperlexia - that of reduced reading comprehension. Thus, in Chapter 4, the main question addressed in Manuscript 2 (referred to as Article 2 from this point forward) was, "Is it possible to improve reading comprehension of preschool children with ASD and hyperlexia at an early age?". The goal was to evaluate a novel intervention that capitalized on the intense interest in early reading to advance reading comprehension. Based on previous assumptions and findings from Article 1 in Chapter 3, that these children were relying on patterns to read, careful consideration was given to word choice included in the intervention. For example, starting with basic Consonant-Vowel-Consonant (CVC) word shapes, as in the word "cat" and moving to longer and more complex word shapes, (e.g., CCVC or CVCC), as in the words "stop" and "fast", to facilitate the reading process and maximally focus attention on the comprehension aspect of reading. Comparing preschoolers with ASD+HPL to preschoolers with ASD-HPL and TD preschoolers permitted assessment of whether the intervention might also impact language abilities in all three groups.

The intervention was based on two theoretical models: the Dual Coding Theory of Literacy (Sadoski, 2005) and the Simple View of Reading (Hoover & Gough, 1990). According to the Dual Coding Theory of Literacy, receptive vocabulary acquisition is facilitated by using the dual code (image plus orthography). In addition, the Simple View of Reading theory states that the product of decoding and linguistic comprehension equals reading comprehension. Given this simple formula, the primary hypothesis was that targeting linguistic comprehension, accelerated via the Dual Coding Theory, would result in a subsequent improvement in both oral language skills in all three groups and reading comprehension skills in the group with ASD+HPL.

Article 2 was the first to examine a strength-based approach to intervention in preschoolers with ASD+HPL. It was also the first to target reading comprehension at such a young age. Currently, reading comprehension is typically targeted in primary school around grade 3. Thus, it was anticipated that promising outcomes might lead to a change in early literacy curriculum. The aims of the study outlined in Article 2 in Chapter 4 are to evaluate the effects of an early, parent supported, tablet-based reading comprehension intervention on the oral language (receptive and expressive) skills and the reading comprehension of preschool children with hyperlexia who could already read, but did not yet have the associated level of reading comprehension.

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Chapter 4: Manuscript 2

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Early Reading Comprehension Intervention for Preschoolers with Autism Spectrum Disorder and Hyperlexia

Abstract

Children with autism spectrum disorder (ASD) and hyperlexia (HPL) have both advanced word reading skills and a reading comprehension disorder, alongside impaired oral language. We developed a unique, parent-supported, tablet-based intervention aiming to improve oral and reading comprehension at the word-, phrase- and sentence-level, for preschoolers with ASD and hyperlexia (ASD+HPL). English-speaking preschoolers (N=30) with ASD+HPL (N=8); ASD without HPL (N=7) and typical development (N=15) underwent a 6-week no-intervention period followed by a 6-week intervention period. Findings revealed a significant increase in reading comprehension scores for the group with ASD+HPL as compared to the other groups (p=.018). Gains were also found for receptive but not expressive language for all groups. Implications for early intervention for preschoolers with ASD+HPL are discussed.

Early Reading Comprehension Intervention for Preschoolers with Autism Spectrum Disorder and Hyperlexia

Introduction

Few would argue that the ultimate goal of reading is to gain meaning from the written text on the page. Through reading we gain knowledge, are entertained, and immersed in different mental worlds. The American author, James Baldwin argued that reading books changed the course of his life by bringing together the "possibilities the books suggested and the impossibilities of the life around me" (Mead & Baldwin, 1971, p. 39). Yet, reading comprehension, the key component to extracting meaning from text, is missing for some children with exceptionalities. Such is the case for those with hyperlexia (HPL), a term first coined by Silberberg and Silberberg (1967), defined as a reading comprehension disorder most commonly associated with Autism Spectrum Disorder (ASD; Ostrolenk et al., 2017). Children with hyperlexia present with poor reading comprehension despite a remarkable strength in early word-level reading and an intense interest in written material (Ostrolenk, et al., 2018). But early success in single-word level reading that lacks associated word-level reading comprehension, referred to as lexico-semantic processing, is largely inefficacious. Reading comprehension has been consistently reported as a significant academic challenge for those with ASD (Randi et al., 2010; Ricketts, et al., 2013).

Unfortunately, children with hyperlexia's word-level reading is not well supported in preschool or kindergarten (Newman et al., 2007). Parents report that early intervention in schools do not target their children's needs, and children with ASD and hyperlexia can be actively discouraged from engaging in reading, their preferred activity (Newman et al., 2007). These factors may contribute to the later decline viewed in reading skills for older children with ASD and hyperlexia. Studies have found that their advanced word reading appears to decline with increasing age to the point where it reaches the normative average at around age 10 years (Grigorenko et al., 2003; Newman et al., 2007). That is, children with both ASD and hyperlexia under the age of five outperform five-year-old children with typical development (TD) as well as 10-year-old children with ASD and hyperlexia for single word reading (Newman et al., 2007). It has been proposed that an increasing disinterest in reading potentially develops in the absence of appropriate support leading to a plateau of reading skills (Newman et al., 2007).

A lack of early reading support is likely not due to any malintent on the part of teachers but more possibly a lack of understanding and training on how to best manage this special interest. Kindergarten and first grade teachers generally focus on early reading instruction that advances single-word-level reading, a skill that children with hyperlexia have already acquired despite their struggles with understanding the meaning of these words. Given that the reading comprehension process is not well understood in the context of development prior to formal schooling, even in typical readers (Randi et al., 2010), it is reasonable that preschool teachers and therapists would not address this unfamiliar profile of early reading skills in young children with ASD, and particularly those with hyperlexia. In fact, the difficulties with lexico-semantic processing may even be overlooked given their superior single-word reading ability.

An absence of early preschool word-level intervention for hyperlexia contrasts starkly with the abundance of research addressing reading comprehension at the sentence and paragraph level with older children with ASD. Several systematic reviews and recent studies have documented strategies such as summarization, graphic organizers, mnemonics, reciprocal teaching and explicit instruction (El Zein et al., 2014; Finnegan & Mazin, 2016; Senokossoff, 2016; Turner et al., 2017) demonstrating varying efficacy at improving sentence- and paragraph-level reading comprehension in older children with hyperlexia. Yet, no study to date has examined the effects of addressing early, word-level reading comprehension abilities in preschool or early school-aged children with ASD and hyperlexia to develop oral and reading comprehension skills during this critical period in the development of language and early literacy skills.

The potential implications of early reading comprehension intervention on both oral language and reading comprehension acquisition for preschool children with ASD and hyperlexia are substantial. Indeed, hyperlexia is frequently accompanied by delayed oral language skills (Grigorenko et al., 2002; Needleman, 1982; Grigorenko et al., 2003) defined as atypical or delayed understanding of spoken language (receptive language) and/or expressive language. A key motive for early intervention is to accelerate development (Landa, 2018) thereby increasing prospects for early academic success and socially appropriate communication exchanges (Landa et al., 2011), in addition to improving oral language skills that are otherwise impaired in ASD but have a critical impact on later academic achievement in reading comprehension (Ricketts et al., 2013). We know from TD children that a strong, bi-directional relationship exists between social and academic competence, whereby poor social skills negatively affect academic achievement; and conversely, weak academic achievement correlates with maladaptive behaviour in the classroom (see Nadeem et. al. 2010 for a review). We also know that children with ASD have both social and academic challenges (Brown & Bebko, 2012; Ricketts et al., 2013) that adversely affect their future. Reduced employment and low post-graduate education rates are a few examples of the associated negative career implications post-high school for this group (Autism Society, 2015). Addressing the gap in services focused on early remediation of reading comprehension and oral language, rather than waiting until middle-or upper-elementary school as is common practice, may help alleviate the negative social, academic and vocational issues linked to ASD.

Theoretical Framework: Simple View of Reading and the Dual Coding Theory of Literacy

The Simple View of Reading (SVR; Hoover & Gough, 1990) has been used to account for the underlying reading comprehension challenges associated with ASD (Ricketts et al., 2013) as well as dyslexia and specific reading comprehension disorders (Catts et al., 2006, 2015). According to the SVR, the product of word decoding, and linguistic comprehension determines reading comprehension. Linguistic comprehension is the ability to understand spoken language, while word decoding is the ability to read words via a whole word (orthographic) and/or a sound-based (phonological) route (National Early Literacy Panel, 2008). Given that children with ASD and hyperlexia demonstrate strong word decoding (Atkin & Lorch, 2006; Cardoso-Martins & Ribeiro da Silva, 2010; Grigorenko et al., 2002; Lamonica et al., 2013; Lin, 2014; Mustika et al., 2014; Newman et al., 2007; O'Connor & Hermelin, 1994; Talero-Gutierrez, 2006), in the presence of weak language comprehension (Carlsson et al., 2013), the resulting poor reading comprehension renders the SVR a fitting explanatory model. In fact, both word recognition and language comprehension predicted reading comprehension in adolescents with ASD (Ricketts et al., 2013). Thus, according to the SVR, targeting children's oral language skills could lead to improved reading comprehension, given their already strong word decoding skills.

Targeting oral language skills at the receptive vocabulary level is an appropriate goal for preschoolers with ASD struggling with oral language challenges (Kover et al., 2013). To do so, the Dual Coding Theory (DCT; Sadoski, 2005) provides a framework for accelerating receptive vocabulary acquisition. The DCT distinguishes between abstract and concrete words, and between two codes: verbal (written words, language) and nonverbal (mental imagery, knowledge). While abstract words, like “love”, require knowledge of semantic relationships and higher order language skills to specify meaning, concrete words, like “tree”, can easily be associated with imagery and thus facilitate earlier acquisition. Hence, according to the DCT, concrete words that have the highest imagery factor are learned more quickly in the presence of the dual code: verbal (written word) plus nonverbal cues (picture). In summary, the DCT provides a scaffolding approach for effectively teaching receptive, concrete vocabulary augmented by orthographic cues to improve linguistic comprehension in children with ASD, while the SVR advocates targeting linguistic comprehension as a means to improving reading comprehension.

In support of the DCT and SVR, a number of studies and evidence-based intervention programs combine pictures and orthography (written words) to develop various aspects of oral and written language. For example, Ricketts et al. (2015) demonstrated that by using orthographic cues to support oral vocabulary learning, TD children, those with specific language impairment and children with ASD could learn novel or nonwords after only one intervention session. A match-to-sample paradigm (matching a picture to a word) was also effective in teaching emergent language skills (receptive and expressive vocabulary) to 3- to 8-year-old children with ASD (Bejnö et al., 2018). While Still et al. (2015) successfully implemented a tablet-based, text-picture matching task to teach derived requesting (i.e., requesting novel objects) for 3- to 12-year-old children with ASD. Programs such as the Picture Exchange Communication System (PECS; Bondy & Frost, 1985) are also based on pictures to foster communication skills for preschool children with ASD, while the Visualizing and Verbalizing program (Bell, 1991) for older children with ASD teaches lexico-semantic processing (rather than simply sight word reading) using images and text combined. Together, these studies suggest that the technique of word-picture matching, incorporating images and orthography, is a viable means of improving vocabulary acquisition and written language comprehension in young children with ASD. At least one study (Still et al., 2015) also demonstrated that computers and tablets can be used to accomplish these goals. To date, no study has examined the effect of targeting early word reading in preschoolers with ASD and hyperlexia to improve oral language (receptive and expressive language) and reading comprehension.

Aims of the Current Study

The current study seeks to harness the strength in early word decoding abilities of preschool children with ASD and hyperlexia to improve both oral language and reading comprehension. We conduct an intervention study to examine oral language (receptive and expressive) and reading comprehension with measurements at three time points. The aim of this study is to evaluate the efficacy of a unique, tablet-based, early reading comprehension intervention program that targets word-picture matching to improve oral language (receptive and expressive) and reading comprehension (lexico-semantic processing) for preschool children with ASD and hyperlexia. We hypothesize that preschool children with ASD and hyperlexia (ASD+HPL) will significantly improve their reading comprehension and oral language skills from a pre-intervention period (normal developmental growth) to post-intervention (following exposure to the intervention), as compared to a TD group and a group with ASD without hyperlexia (ASD-HPL).

Method

Participants

We have previously discussed the participants in this study (recruitment, ascertainment criteria, inclusionary and exclusionary criteria, and baseline measures) and their associated characteristics in-depth in an earlier study (Macdonald et al., 2020) [Article 1] that is summarized here. A total of 32 participant pairs (parents and their preschool children between the ages of 3 years, and 5 years and 11 months) were included in the intervention study, *N*=16 with typical development (TD) and *N*=16 with ASD. However, two participants (one from the TD group and one from the group with ASD) did not complete the intervention requirements and were removed from the study and their subsequent data were not included in analysis. Thus, *N*=15 were included in the TD group and *N*=15 were included in the group with ASD (see Table 1). The group with ASD was further divided into those with hyperlexia (ASD+HPL), *N*=8 and those without hyperlexia (ASD-HPL), *N*= 7.

Table 1

Age, FSIQ, VIQ, word reading, ASD and hyperlexia symptoms for all groups before the reading intervention

	ASD+HPL (*N*=8)		ASD-HPL (*N*=7)		All ASD (*N*=15)[d]		TD (*N*=15)	
	M(SD)	Range	*M(SD)*	Range	*M(SD)*	Range	*M(SD)*	Range
Age (months)	53(9)	36-65	58(11)	41-70	55(10)	36-70	49(8)	38-61
WPPSI-IV^{CDN} FSIQ[a**c**]	69(8)	58-78	82(10)	67-94	75 (11)	58-94	87 (12)	68-112
WPPSI-IV^{CDN} VIQ[a**b*c**]	70(9)	58-85	88(13)	67-102	79(14)	58-102	95 (12)	71-111
WJ-IV-ACH LWI[a**b**c**]	121(14)	98-138	93(17)	72-110	95(14)	73-119	108(21)	72-138
WJ-IV-ACH PC	96(26)	51-123	91(19)	60-115	93(22)	51-123	98(10)	82-113
ADOS-2 Comparison Score	7(2)	4-10	6(2)	4-9	7 (2)	4-10	----	----
SRS-2P[c**]	64(9)[2]	54-78	74(7)	63-84	69 (9)	54-84	46 (6)	39-60[e]
SCQ-L[c**]	13(6)[2]	3-20	18(5)	12-24	16 (6)	3-24	8 (9)	1-29[f]

Note: Data reported in this table (excluding the data on the WJ-IV-ACH PC) was first reported in Macdonald et al., 2020 [Article 1]
Significant difference *p<.05, ** p<.01 between: (a)ASD+HPL vs TD, (b)ASD+HPL vs ASD-HPL, (c)ASD vs TD
WPPSI-IV^{CDN} FSIQ and VIQ; WJ-IV-ACH LWI and PC (standard scores); SRS-2P (t-scores)

[d]All ASD (n=15): ASD+HPL (n=8) plus ASD-HPL (n=7)
[e]All participants in the TD group had scores at or under the cut-off score for ASD (*t*-score≥60) on the SRS-2P.
[f]Two participants in the TD group had scores above the cut-off (>10) on the SCQ-L. Both these participants had scores below the cut-off score for ASD (*t*-score≥60) on the SRS-2P. The range of SCQ-L scores in the TD group without two outliers is from 1-10

There were no significant differences between the groups for age (see Table 1) nor family income. However, the groups did differ significantly for gender. Consistent with previous studies of ASD+HPL (Grigorenko et al., 2002; Newman et al., 2007) there were significantly more males in the sample with ASD (93%). The opposite was true for the TD group, composed of significantly more females (73%). Using chi-square analysis, there was no statistically significant difference for paternal education, but all three groups differed significantly on maternal education (p=.04).

Study and Subgroup Inclusion and ASD Symptoms

All children in the group with ASD had a diagnosis provided by a registered clinical psychologist, paediatrician or psychiatrist, in addition to meeting criteria on the Autism Diagnostic Observation Schedule, 2nd edition (ADOS-2; Lord & Rutter, 2012). ADOS-2 Comparison scores were prioritized for inclusion with a range of 4-10 (low to high severity) required. In addition, the Social Communication Questionnaire – Lifetime (SCQ-L; Rutter, Bailey, & Lord, 2003) and the Social Responsiveness Scale – Second Edition – Preschool Version (SRS-2P; Constantino, 2012) provided secondary inclusionary measures for the ASD group and exclusionary measures for the TD group. There was a statistically significant group difference (ASD vs TD) for scores on the SCQ-L, p<.006, and the SRS-2P, p<.001, in favour of higher scores for the group with ASD (see Table 1) indicating clinically significant symptomology associated with ASD. Participants with ASD+HPL (N=3) who did not meet criteria on either the SRS-2P and/or the SCQ-L were included in the group if they met criteria on the ADOS-2, ADOS-2 comparison score and hyperlexia inclusion. All children with ASD and

TD children were included if parents confirmed that their child spoke English at home daily with at least one parent since birth, had an interest in letters, words, and/or an ability to read words, to ensure that children would be interested in the intervention. Children with a history of receiving formal reading instruction were excluded. No child in either group had a documented history of a failed hearing test, nor symptoms of hearing loss during the study.

Hyperlexia Inclusion

As previously discussed in Macdonald et al. (2020) [Article 1], we based inclusion in the ASD hyperlexia group (ASD+HPL) on five criteria according to Needleman (1982) as follows: (a) early word reading before age 5 years, (b) a Full Scale IQ (FSIQ) below 78 as measured on the Weschler Preschool and Primary Scale of Intelligence – Fourth Edition: Canadian, (WPPSI-IV^{CDN}; Wechsler, 2012), (c) a Verbal IQ (VIQ) below 85, as measured on the VIQ scale of the WPPSI-IV^{CDN}, (d) single word reading 2 standard deviations above VIQ and FSIQ as measured on the Letter-Word Identification (LWI) subtest of the Woodcock Johnson Test of Achievement – Fourth edition (WJ-IV-ACH; Schrank, Mather, & McGrew, 2014), and (e) poor reading comprehension relative to word reading as measured on the Passage Comprehension subtest (PC) of the WJ-IV-ACH. Of the eight participants in the group with ASD+HPL, seven met all five criteria and one met four criteria. The remaining 7 participants with ASD were assigned to the ASD-HPL group.

Single Word Reading, Reading Comprehension, FSIQ and VIQ

As previously shown in Macdonald et al. (2020) [Article 1], (see Table 1), statistically significant differences were found between all three groups for single word reading on the WJ-IV-ACH LWI, $F(2,23)=10.46$, $p<.001$, with significantly higher mean word reading scores for the group with ASD+HPL as compared to both the TD group ($p=.001$) and the group with ASD-

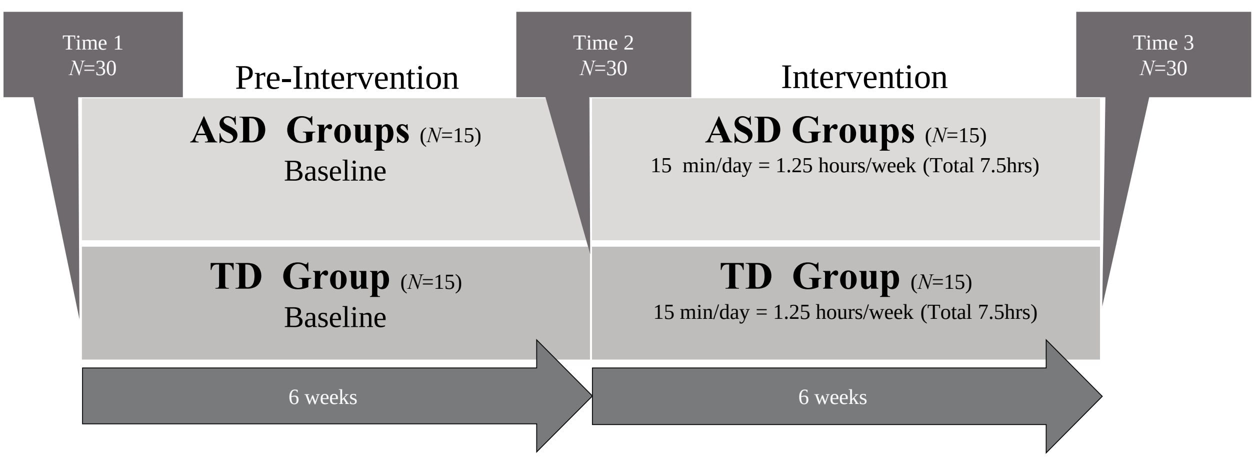
HPL group (*p*=.004). In addition, there were statistically significant group differences for both FSIQ, $F(2,27)=7.43$, $p=.003$, and VIQ, $F(2, 27)=11.83$, $p<.001$, with significantly higher mean FSIQ ($p=.002$) and VIQ ($p<.001$) for the TD group as compared to the group with ASD+HPL group, and higher VIQ for the group with ASD-HPL as compared to the ASD+HPL group ($p=.021$). It is worth noting that the hyperlexia identification criteria included lower VIQ and FSIQ and thus, it is not surprising that the group with ASD+HPL would have poorer scores in these domains. In contrast, there were no significant differences among groups for the distribution of scores on reading comprehension (symbol and text representation) on the WJ-IV-ACH PC before the intervention.

Design

In this repeated-measures A-B design (see Figure 1), parent and child dyads with ASD (*N*=15), and TD (*N*=15) completed baseline measures at Time 1.

Figure 1

Study Design



Note: ASD Groups: ASD+HPL (*N*=8) and ASD-HPL (*N*=7)

Following Time 1 assessment, parent-child dyads underwent a pre-intervention period for 6 weeks (from Time 1-2). This initial pre-intervention period was designed to serve as a measure of normal developmental growth that occurs in the absence of intervention, representing normal maturation. Thus, each participant served as their own pre-intervention control. During this period, parents were instructed to interact with their child as they normally did at home. After this six-week period, children were reassessed (Time 2). The tablet-based reading intervention period then lasted 6 weeks (from Time 2-3) in the participants' homes where parents supported their child's use of the tablet intervention for 15 minutes per day, five days per week, for a total of 7.5 hrs of intervention (See Figure 1). Parents were instructed to conduct the intervention with their child at a time that worked for their daily schedules. Parents were asked to sit with their child during the intervention time and interact as they would when reading a book, as well as to provide praise and encouragement. Subsequent to the intervention period, children completed Time 3 assessments (post-intervention).

Intervention

The first author and a team of professional developers at Brandjaws (Mansoor, 2020) through UpWork (Karamanlakis & Tsatalos, 2015) designed and built the reading comprehension intervention application on an iOS platform with a web-based backend to collect data. The intervention implemented a forced-choice, two-option, text-to-picture matching activity (see Figure 2 and Figure B1) with stimuli that consisted of 145 concrete nouns, 100 verbs, 48 adjectives, 24 concepts and 11 pronouns organized from early to later developing words, based on the *MacArthur-Bates Communicative Development Inventories Wordbank* (Frank et al., 2017). All children began at the first item, with concrete nouns of short word shapes (e.g., Consonant Vowel Consonant; CVC), like *cat*, and progressed to longer noun word shapes (e.g., CCVC or CVCC) like *slip* or *park*. There were three levels: word-, phrase-, sentence-picture matching tasks, and two phases: Oral Language and Reading Comprehension (see Figure B2 and B3).

Figure 2

Sequence of screen displays with the tablet-based reading comprehension and oral language intervention.

The screen display, in both the Oral Language and Reading Comprehension phases, was identical; each presented a written word (e.g., bed; see Figure 2 and Figure B1). The only difference between the two phases was audio (the spoken word) accompanied the written word along with an ear icon in the Oral Language phase, while no accompanying audio and a book icon appeared during the Reading Comprehension phase. The child heard the verbal prompt (via a synthesized iPad female voice) to "Touch the word". There was an available option in the settings to replace the verbal prompt with a nonverbal green arrow pointing to the word. In the

Oral Language phase, the participant heard the oral word when touching the written word, e.g., the participant heard and saw the word *bed* simultaneously. In each phase, after the child touched the word, two images appeared on the screen (the corresponding image and a distractor image) above the same written word (see Figure 2 and Figure B2). The child then heard the verbal prompt “Touch the picture that goes with the word” or no prompt (if verbal prompting was turned off in the settings). The child then chose the picture that corresponded to the written and auditory word in the Oral Language phase or to the written word in the Reading Comprehension phase by touching the image.

When a child touched the correct picture, they heard a rewarding sound (ascending tones; differential reinforcement) or verbal reinforcement (That’s right! or well done!) depending on the chosen settings. The correct image was then highlighted in green, and the ‘next’ arrow appeared, which the child selected to advance to the next word. When the child chose the incorrect word (distractor), a corresponding “incorrect” sound (descending tones) or verbal prompt option, “Try again” played and the same two pictures in the same orientation were repeated. If the child chose the incorrect image a second time, that image faded and only the correct image appeared for selection. On a new trial, the target image remained the same with the position randomly varying between left and right and the distractor image randomly alternating between two different images. Each trial always ended with the child choosing the correct picture and receiving positive verbal or auditory reinforcement.

The intervention incorporated a number of reward systems to maintain motivation. For example, during the intervention a small sunshine icon appeared every 3 minutes for a total of 5 icons over the 15-minute session (see Figure 2). At the end of each 15-minute session, a link to PBS kids’ games appeared. However, parents were also encouraged to provide their own form of reward if they did not want their child to play iPad games. Finally, at the completion of 5 days of intervention a message appeared on the screen with a present icon congratulating the child on their work and letting them know they had earned a gift. It was left up to the discretion of the parent as to whether the child would choose his/her own gift, or whether the parent would provide them with one. Gifts provided to parents for their child included pens, colouring sets, bubbles, toy cars, etc. This was repeated each week for the 6-week period (for a total of 6 gifts throughout the intervention period).

Data Collection

We used standardized tests at Times 1, 2, 3 to assess outcome measures of reading comprehension (symbol and text representation) and oral language (receptive and expressive) during the intervention period (from Time 2-3), as compared to a pre-intervention period (from Time 1-2). Baseline measures, including the ADOS-2, SRS-2P, SCQ-L, WPPSI-IV^{CDN} and the WJ-IV-ACH LWI, were collected at Time 1.

Standardized Outcome Measures (Time 1, 2 and 3)

Reading Comprehension. The primary outcome measure used to assess reading comprehension (symbol and text representation) was the Passage Comprehension subtest of the Woodcock Johnson Test of Achievement – Fourth edition (WJ-IV-ACH PC; Schrank, et al., 2014). The WJ-IV-ACH PC subtest measures symbol, single word and sentence comprehension. Since we also assessed TD preschoolers who were not yet reading, we refer to this task as symbol and text representation (see Table 1). Versions B and C of the WJ-IV-ACH were randomly assigned to ensure counterbalance of the presentation order across Time 1, 2 and 3. Raw scores for the WJ-IV-ACH subtests were used for consistency in analysis given that standard scores are not available for children younger than 5 years of age on the B version of the OWLS-II Listening Comprehension and Oral Expression Scales.

Oral Language. The primary outcome measures used to assess oral language skills were the Oral and Written Language Scales (OWLS-II; Carrow-Woolfolk, 2011) Listening Comprehension Scale (OWLS-II LC) and the Oral Expression (OWLS-II OE) Scale, administered to evaluate receptive and expressive language skills respectively. Versions A and B of the OWLS-II were also randomly assigned to ensure counterbalance of the presentation order across Time 1, 2 and 3. Raw scores for the OWLS-II subtests were used in analysis as noted.

Web-based Data Collection

A web-based database securely and anonymously collected information on performance and usage including date, number of sessions spent on the intervention on a daily basis, number of correct and incorrect words choices, total daily score on each of the Oral Language and Reading Comprehension phases of intervention and final level attained (see Table B4). Chi square analysis demonstrated a significant difference for the group with ASD+HPL for final level attained ($p<.05$), with no participants finishing at the word level (see Table B4). There was no significant difference between the three groups in mean number of sessions spent on the intervention ($p=.074$; ASD+HPL, $M=22$; ASD-HPL, $M=21$; TD, $M=16$).

Compliance and/or Intervention Fidelity. Participants were required to complete the intervention five days per week. Parents were sent friendly reminders via email or contacted directly if the weekly goals were not met. For example, any participant who missed three consecutive days or who failed to complete the entire 15 minutes on three consecutive days, was contacted to offer assistance or troubleshoot with the family. If a child did not complete five days in any two weeks of the study, they were removed from the study; this was the case for two participants who were not included in the final sample of 30 participants as noted.

Settings

Assessment activities were conducted either at McGill University or a clinical psychology office. Occasionally, assessments were conducted in the participants' homes. Some children were able to complete all Time 1 measures during the same session, while others required two separate sessions. Child participants received a small gift following each assessment session. All intervention sessions were conducted in the families' homes by the parents after receiving instructions and a demonstration with their child. Parents received a $25 gift card for their participation. Families who did not own an iPad were lent one for the 6-week intervention period.

Statistical Analyses

Data were coded to ensure anonymity and entered into SPSS statistical software version 25 and analysed in version 26. We aimed to evaluate whether the intervention significantly improved reading comprehension and oral language (receptive and expressive) scores from the initial pre-intervention period (Time 1-2) as compared to the intervention period (Time 2-3). Data screening was performed prior to analysis. Assumption checks for the two-way mixed ANOVA revealed a number of extreme outliers in the data, as evidenced by inspection of box plots. Floor effects were noted in the TD group on the WJ-IV-ACH Passage Comprehension subtest. Normality was assessed using the Shapiro-Wilk test, where a significance of $p<.001$ was used to identify data that were not normally distributed. The assumption of homogeneity of variances was violated for the symbol and text representation variable (assessed via WJ-IV-ACH PC) for Time 1 and 2. The assumption of homogeneity of covariances was violated as evidenced by a significant Box's test (*p*=.010). Given the violations of assumptions we elected to run Friedman's nonparametric repeated measures ANOVA separately for the three groups for the reading comprehension (symbol and text representation) variable. We used the Kruskal-Wallis tests for the group comparisons at each time point. We also examined the improvement in scores from Time 2-3 using linear regression to obtain standardized residuals and compared these between groups using the Independent samples Mann-Whitney U test. When all assumptions were met, we used a two-way mixed ANOVA to assess if there was a two-way interaction among the between-subjects factor (groups: ASD+HPL, ASD-HPL, TD) and the within-subjects factor (Time 1, 2, and 3) on the dependent variables (receptive and expressive language).

Results

Reading Comprehension

Symbol and text representation

Friedman's nonparametric repeated measures test revealed a statistically significant difference in the distribution of scores for symbol and text representation across the three different time points, $\chi^2(2, 30)=16.796$, $p<.001$, $\eta^2 = 0.28$ (see Table 2, Figure 3), as measured on the WJ-IV-ACH PC subtest. Pairwise comparisons with Bonferroni correction indicated that these differences reflected no significant difference from Time 1-2, *p*=1.00, but significant improvements in performance from Time 1-3, *p*=.002, and from Time 2-3, *p*=.007. These results indicate that scores on reading comprehension (symbol and text representation) were significantly higher following the intervention period (at Time 3) as compared to the pre-intervention period (at Time 1 and at Time 2) for all groups.

The Kruskal-Wallis test was run to examine differences between the three groups at Times 2 and 3. The distributions of symbol and text representation scores demonstrated significantly different scores among the three groups at Time 3 only $\chi^2(2,30)=7.783$, $p=.020$, $\eta^2=.14$, but not at Time 2, $\chi^2(2,30)=4.203$, $p=.122$. Pairwise comparisons of group with Bonferroni correction indicated that these differences were between the TD group and the group with ASD+HPL, $p=.017$, but not between the TD group and the group with ASD-HPL, $p=1.00$, nor between the groups with ASD+HPL and ASD-HPL, $p=.208$ (see Figure 3, Table 2). Thus, the group with ASD+HPL had significantly higher mean scores in symbol and text representation post-intervention (Time 3) as compared to the TD group, but not compared to the group with ASD-HPL.

To further examine improvement in scores noted from Time 2-3 for all groups, and higher scores at Time 3 for the ASD+HPL group compared to the TD group, we regressed the change in scores from Time 1-2 (Δ 1-2) out of the change in scores from Time 2-3 (Δ 2-3) for both groups and compared the unstandardized residuals across groups using an Independent-Samples Mann-Whitney U test. Results showed again that the group with ASD+HPL (mean rank = 16.31) had significantly greater gains in scores from Time 2-3 as compared to the TD group (mean rank =9.87), $U=94.5$, $p=.023$, $\eta^2=.23$ when controlling for the change in scores from Time 1-2 scores. These results indicate greater improvement in reading comprehension (grasp of symbol and text representation) associated with the intervention from Time 2-3 for the ASD+HPL group compared to the TD group.

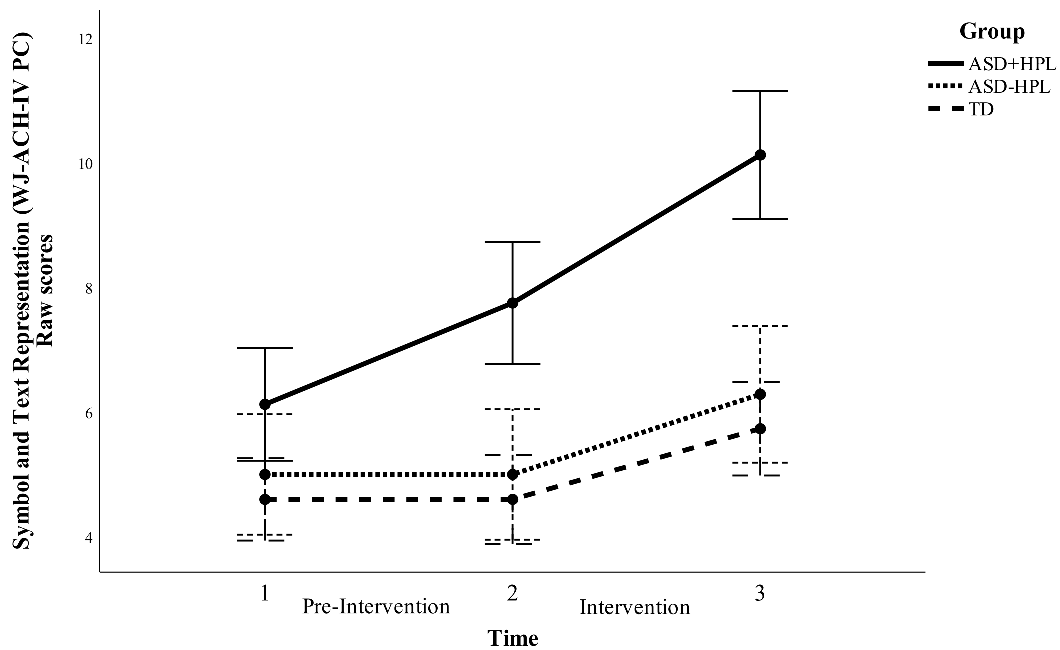
Table 2

Mean Raw Scores on Outcome Measures at Time Points 1, 2, and 3 for the ASD+HPL, ASD-HPL, and TD groups

	ASD+HPL			**ASD-HPL**			**TD**		
	Time *M(SD)*			Time *M(SD)*			Time *M(SD)*		
Variable	1	2	3	1	2	3	1	2	3
Reading Comprehension WJ-IV ACH PC	6.12 (4)	7.75(5)	10.13(4)	5.0(3)	5.0(1)	6.28(2)	4.6(1)	4.6(2)	5.73(2)
Receptive Language OWLS-II LC	23.62(10)	23.25(14)	32.5(17)	36.14(18)	36.86(12)	44.57(17)	36.2(21)	41.13(20)	45.00(21)
Expressive Language OWLS-II OE	8.88(5)	14.25(6)	13.0(7)	21.71(10)	25.0(8)	28.43(11)	23.6(12)	26.73(13)	28.73(12)

Figure 3

Mean reading comprehension (symbol and text representation) a time points 1, 2, and 3 for ASD+HPL, ASD-HPL and TD groups (error bars = +/- 1 SE)



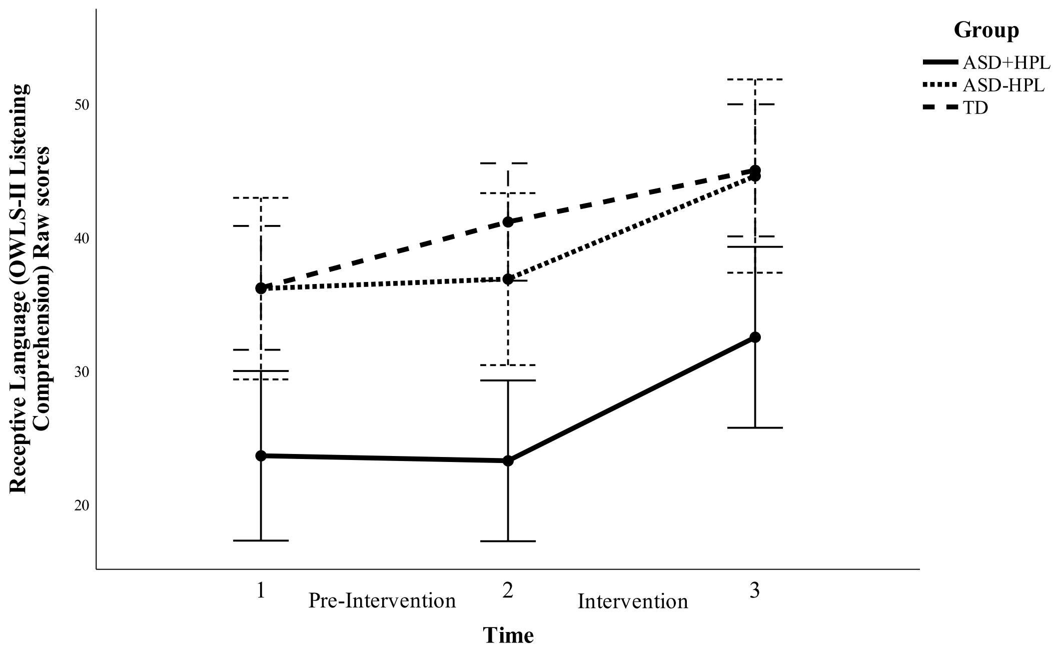
Oral Language

Receptive Language

A two-way mixed ANOVA was run for receptive language as assessed on the OWLS-II Listening Comprehension Scale (see Figure 4, Table 2). There was no significant group x time interaction for receptive language, $F(4, 54)=.725$, $p=.579$, $\eta^2=.051$. The main effect of group was not statistically significant, $F(2,27)=1.887$, $p=.171$, $\eta^2=.123$. However, the main effect of time showed a statistically significant difference in mean receptive language at the different time points, $F(2,54)=14.138$, $p<.001$, $\eta^2=.344$. Pairwise comparisons with Bonferroni correction revealed no significant difference from Time 1-2, $p=.929$, with significant improvement in performance from Time 1-3, $p<.001$, and from Time 2-3, $p=.002$. This finding indicates that the mean receptive language scores were higher post-intervention (at Time 3) as compared to pre-intervention (at Time 1 and Time 2) and that this was the case for all three groups taken together.

Figure 4

Mean receptive language measured on the OWLS-II Listening Comprehension Scale at time points 1, 2 and 3 for the ASD+HPL, ASD-HPL and TD groups (error bars = +/- 1 SE)



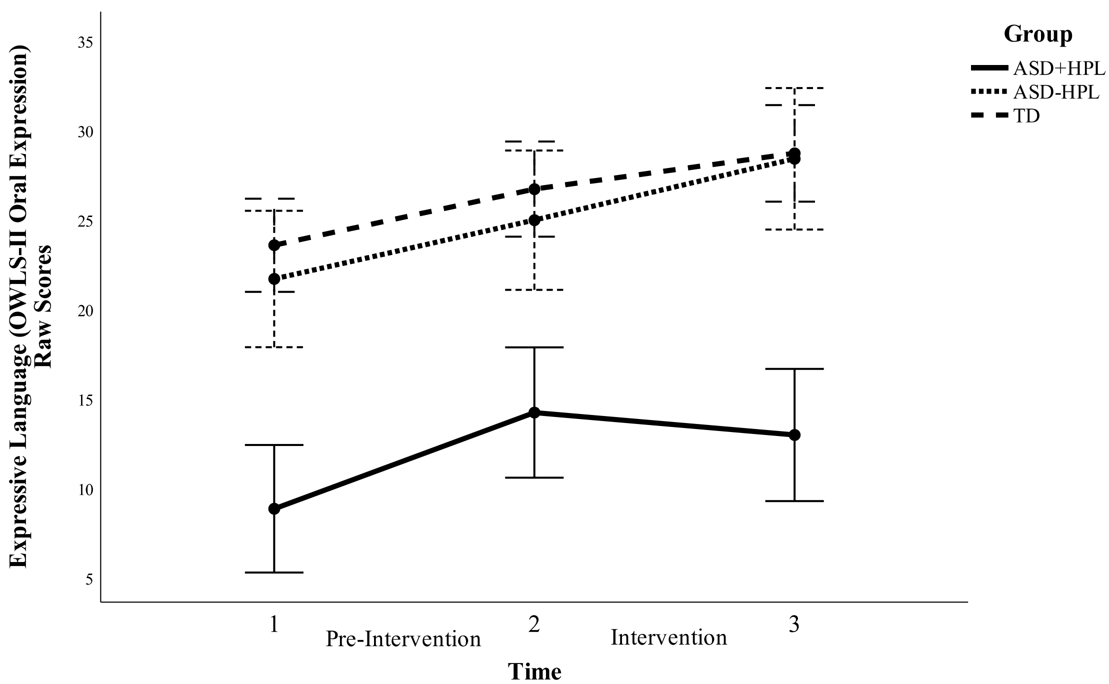
Expressive Language

A two-way mixed ANOVA was conducted to examine expressive language as measured on the OWLS Oral Expression Scale. There was no significant group x time interaction for expressive language, $F(4,54)=1.182$, $p=.329$, $\eta^2=.080$ (see Figure 5, Table 2). There was a significant main effect of group, $F(2,27)=5.773$, $p=.008$, $\eta^2=.300$. Pairwise comparisons with Bonferroni correction revealed a significant difference between the TD group and the group with ASD+HPL, $p=.008$, with the TD group demonstrating higher mean expressive language scores throughout the pre-intervention and intervention periods. The difference between the groups with

ASD+HPL and ASD-HPL approached significance, *p*=.053, revealing the group with ASD-HPL to trend towards having higher mean scores for expressive language throughout the pre-intervention and intervention periods as compared to the group with ASD+HPL. The main effect of time was also significant, $F(2,54)=20.208$, $p<.001$, $\eta^2 = .428$. Pairwise comparisons with Bonferroni correction indicated significant differences in oral expression from Time 1-2, *p*<.001 and from Time 1-3, *p*<.001, but not from Time 2-3, *p*=.414. Thus, the intervention did not appear to significantly affect change in oral expression skills in the group with ASD+HPL nor did it affect change in oral expression differently from one group to another.

Figure 5

Mean expressive language measured on the OWLS-II Oral Expression Scale at time points 1, 2 and 3 for the ASD+HPL, ASD-HPL and TD groups (error bars = +/- 1 SE)



Discussion

This study is the first to report on a successful early intervention for preschoolers with ASD+HPL targeting their strength in early word reading to improve areas of challenge in word-, phrase- and sentence-level reading comprehension and oral language skills. Results demonstrated that significant improvements in reading comprehension (symbol and text-representation) and receptive language can be achieved at the preschool level with very young children with ASD+HPL. This study adds to other successful, parent-supported, tablet-based language interventions for young children with ASD (Dunn et al., 2017; Still, et al., 2015), in addition to contributing to previous findings of existing intervention programs that use visuals to support language development (Bell, 1991; Bondy & Frost, 1985) and studies that have effectively used word-picture matching (Bejnö, et al., 2018; Still, et al., 2015) and orthography (Ricketts et al., 2009, 2015) to teach language skills to children with ASD.

Our hypothesis stated that preschoolers with ASD+HPL would improve their reading comprehension and oral language skills from pre-intervention (from Time 1-2) to post-intervention (from Time 2-3) as compared to the two comparison groups (TD and ASD-HPL). Our findings support this hypothesis for reading comprehension for the group with ASD+HPL, whose reading comprehension scores significantly increased post-intervention compared to pre-intervention on the WJ-IV-ACH PC subtest measuring symbol and text representation, as compared to the TD group (see Figure 3). Thus, a 6-week duration of reading comprehension intervention accelerated reading comprehension acquisition in early readers with ASD+HPL (from Time 2-3) as compared to their normal growth trajectory prior to intervention (from Time 1-2) and as compared to a TD group. Clearly, these results suggest that there is no need to delay intervention until later primary school to affect change in reading comprehension.

For oral language skills, our hypothesis proved to be correct for all groups in that there were significantly higher receptive language scores following the intervention period as opposed to the pre-intervention period for all groups taken together. Increased oral language skills post-intervention support and extend those of Ricketts et al. (2009, 2015) by showing that orthography plus imagery can improve receptive language, not just oral vocabulary, in preschool children with ASD. Moreover, given the strong link between vocabulary development and reading success (Perfetti, Yang, & Schmalhofer, 2008), with a correlation of 0.81 between vocabulary knowledge and reading comprehension in the general population of school-age children (Biemiller, 2003), improving receptive vocabulary is key to accelerating reading comprehension (Carlson et al., 2013).

The findings of this intervention also strengthen the application of the Simple View of Reading (SVR) to reading comprehension intervention for preschoolers with ASD+HPL. The notion of targeting linguistic comprehension to improve reading comprehension when word decoding is intact has been supported by the results of this intervention. Similarly, the improvements in receptive language comprehension also provide support for the use of the Dual Coding Theory (DCT) as an effective strategy, to not only teach vocabulary to preschoolers with ASD+HPL, but to support reading comprehension. By incorporating concrete, high imagery vocabulary terms and their written correlates as targets, Sadoski (2013) maintains that learning and semantic memory are enhanced, similar to a mnemonic effect, via the connection of verbal (visual language – writing) modalities and nonverbal (mental representations of an object) modalities. “Without the activation of mental representations, no meaning can be present; the potential in the cognitive system lies dormant.” (Sadoski, 2013, p. 50).

For expressive language skills, the ASD+HPL group's gains following the intervention period were not significantly different from the gains made by the other groups (see Figure 5). Although we had hypothesized that all aspects of oral language would be favourably impacted by the intervention, this was not the case for expressive language skills. Yet, these results may shed light on the nature of language acquisition in children with ASD. Post-intervention, all groups showed a similar pattern of acquisition, with gains in receptive language but not expressive language. In TD children, receptive language typically precedes expressive language acquisition (Lahey, 1998; Carrow-Woolfolk, 2011). While it is commonly held that preschool children with ASD demonstrate more severe deficits in receptive vocabulary (Kover et al., 2013) than expressive language (Davidson & Ellis Weismer, 2014), a recent meta-analysis of 74 studies did not support this finding, showing instead a similar level of impairment across both receptive and expressive language (Kwok et al., 2015).

Our findings for young children with ASD+HPL are consistent with these meta-analytic findings supporting weaker development in both language domains as compared to control groups, and contrast with previous studies of ASD+HPL where language was on par with non-hyperlexic and TD groups (Grigorenko et al., 2002; Newman et al., 2007). Thus, higher receptive language gains in our study, as compared to the poorer outcomes in expressive language profile across all groups may reflect a more typical rather than atypical language acquisition pattern for children with ASD. However, in light of the known heterogeneity in ASD (Baio et al., 2018) and in ASD+HPL for language acquisition across studies (Grigorenko et al., 2002; Newman et al., 2007), in combination with the variation in criteria used to define hyperlexia (Ostrolenk et al., 2018), more studies are required to examine this pattern of linguistic development in preschoolers with ASD+HPL.

Additional Considerations

There are a number of ways that gains in reading comprehension may have been fostered by this intervention for the group with ASD+HPL. Firstly, parental support during the intervention may have been a factor. A recent review of early intervention for children under five years old, at-risk or diagnosed with ASD, Landa (2018) concluded that "equipping parents to implement development-enhancing strategies while engaged with their child is a vital intervention component" (pp. 34-35). Another possible contributing factor may have been the use of a tablet. The tablet's touch screen afforded the use of a simple pointing response. It has been noted that an abstract gesture (touching the screen) tied to the underlying concept of the task (in this case identifying an object by pointing) that avoids irrelevant features of the real-world task, can result in a greater degree of generalization (McEwen & Dubé, 2017). Thus, parent support paired with a tablet platform and a simple pointing response may each have contributed uniquely to support gains made during the intervention. Future studies should further examine these factors in designing tablet-based interventions for children with ASD as well as investigate their associated impact on generalization.

Additionally, contingent responses in the form of touching the screen and hearing the word or touching a "next" button to proceed to the next page, permitted child-control of the device or activity in contrast to adult-control. Child-control has been shown to improve TD preschool children's visual attention and interest while playing an early reading computer game (Calvert et al., 2005). These authors also found that boys, and those with better reading skills, made more attempts to control the game. This is particularly relevant given the higher number of boys in our ASD+HPL sample (also reflected in the general population of children with ASD; Centers for Disease Control and Prevention, 2014), and their accompanying strong reading skills.

Thus, future studies may benefit from exploring these features in greater depth in early interventions with children with ASD.

Clinical and Educational Implications

The positive effects of early intervention for children with ASD are well documented (see Landa, 2018 for a current review of the literature). Our study not only demonstrated significant benefits for the children, but also suggests additional benefits for parents, therapists and teachers of young children with ASD+HPL. For example, parents can successfully implement this intervention at home at the first signs of hyperlexia. Given the rising prevalence of ASD (Baio et al., 2018), the lengthy wait times for early intervention of upwards of a year (Brian, et al., 2019; Gordon-Lipkin et al., 2016), and the current movement to include children with ASD into mainstream elementary schools (United Nations, 2016; Vakil et al., 2009), addressing early reading skills of preschool children with ASD and hyperlexia in a timely manner is essential. Clearly, there is a need for immediately accessible, simple-to-administer, convenient, evidence-based measures to fill the gap in early service provision.

In addition, daily intervention supported by a parent can closely follow a distributed practice learning schedule (short, daily sessions with frequent, regular practice) that has been shown to be a superior learning paradigm over massed practice (e.g., conducting all trials on one day) for preschool and school-age children with ASD (Haq et al., 2015). As the cost of daily therapy can be prohibitive for families or access to therapy can be limited in remote areas, a tablet-based format offers a relatively cost-effective method of service delivery to preschoolers with ASD while awaiting public services. Finally, encouraging and providing support for their child's special interest for written material early on may help alleviate the later decline found in reading interests and skills around 10 years of age (Grigorenko et al., 2002; Newman, et al., 2007) while simultaneously accelerating growth in both oral and reading comprehension.

For educators and clinicians, findings of this study also demonstrate that there is no reason to wait until late primary school to begin working on reading comprehension. Teachers, learning specialist, and clinicians can easily recommend or implement this program in kindergarten as part of the general curriculum, or in a clinical or special education setting, with no additional training required and minimal support. Similarly, clinicians can include this intervention as an adjunct to their intervention that can easily be carried over at home. This has significant implications for children with ASD+HPL in both clinical and academic settings, as oral language and reading comprehension are essential components of most academic subjects and are primary targets for clinical therapy. Consequently, we argue that there is a lack of evidence to support postponing reading comprehension therapy until the child is older in favour of sentence- and paragraph-level comprehension therapy, but rather early intervention of lexico-semantic processing (word-level comprehension) at the first signs of hyperlexia, is highly advantageous for both reading and oral language development. Early interventions, such as this one, are also a significant step toward establishing best practices for young children with ASD+HPL. In an earlier study, Macdonald et al. (2020) [Article 1] suggest that preschoolers with ASD+HPL do not likely benefit from the same early reading instructional model as their TD peers given their alternative, non-phonological approach to word reading. The current study indicates that focusing on vocabulary development and reading comprehension are viable, alternate early reading intervention routes that can represent best practices for preschoolers with ASD+HPL.

Social and Cognitive Implications

There are potential long term academic, cognitive and social benefits to targeting language and reading comprehension early on. Stanovich (1986) maintained that a reciprocal relationship between vocabulary development and reading comprehension continuously fosters joint advancement in both oral language and reading over the course of a child's lifetime. That is, the more the child reads, the greater their receptive vocabulary growth, and thus the higher their oral comprehension and reading comprehension levels. In support, a large, longitudinal study by Sparks, Patton, and Murdoch, (2014) showed that early reading levels in grade 1 predicted reading comprehension, linguistic ability and general knowledge subsequently in grade 10. Thus, early intervention, such as learning to read for meaning early in life, can stimulate a self-perpetuating system of increasing growth in both written and oral language development (Landa & Kalb, 2012).

This continuous cycle of reciprocal growth may also apply to receptive language, social interaction and reading comprehension. In a study of preschoolers with ASD aged 2.6 years to 6.5 years, Delinicolas and Young (2007) confirmed that both language (receptive and expressive) and social development were significantly correlated with joint attention (both initiating pointing and following a point). As well, Ricketts et al. (2013) found that social impairments in adolescents with ASD were associated with reading comprehension difficulties. Thus, a reciprocal relationship between language development, early social development and reading comprehension may be fostered by early interventions such as the one presented in this study.

Conclusion and Future Directions

In summary, this early reading comprehension intervention represents a paradigm shift in the current method of teaching reading comprehension. Instruction has typically been delivered in later grade school and beyond at the sentence- and paragraph-level. The current study suggests that early intervention for lexico-semantic processing, beginning at the single-word level, as opposed to paragraph-level, might offer a valuable language learning opportunity for a child with ASD and support early language and academic skills. Our study is the first to report on a successful, strength-based, single-word approach targeting reading comprehension in very young children with ASD+HPL, although future studies with larger sample sizes are required to confirm these benefits. Early reading and language comprehension intervention holds the potential to mitigate the social and academic challenges that children with ASD face (Brown & Bebko, 2012; Ricketts et al., 2013). Improving employment and post-graduate education rates and reversing the negative career and academic outcomes post-high school that have been documented for this group (Autism Society, 2015) are key motives in developing earlier interventions to improve academic and functional skills including reading comprehension. Ultimately, and most importantly, early reading-based intervention may permit children with ASD+HPL to apply their strength in, and love of reading to develop language and reading comprehension in parallel so that reading becomes a rich and meaningful experience, one that can positively influence the personal, academic and professional course of their lives.

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Appendix B

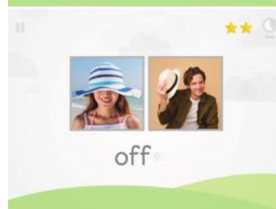
Figure B1

Examples of verbs, adjectives, pronouns, concepts, phrases and sentences used in the intervention

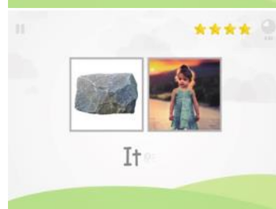
Verb

Adjective

Concept



Pronoun



Phrase

Sentence

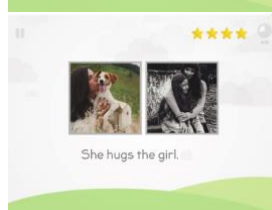


Figure B2

Successful or Unsuccessful Acquisition of a Target Word in 2/3 Trials in a Learning Opportunity: Three Possible Avenues

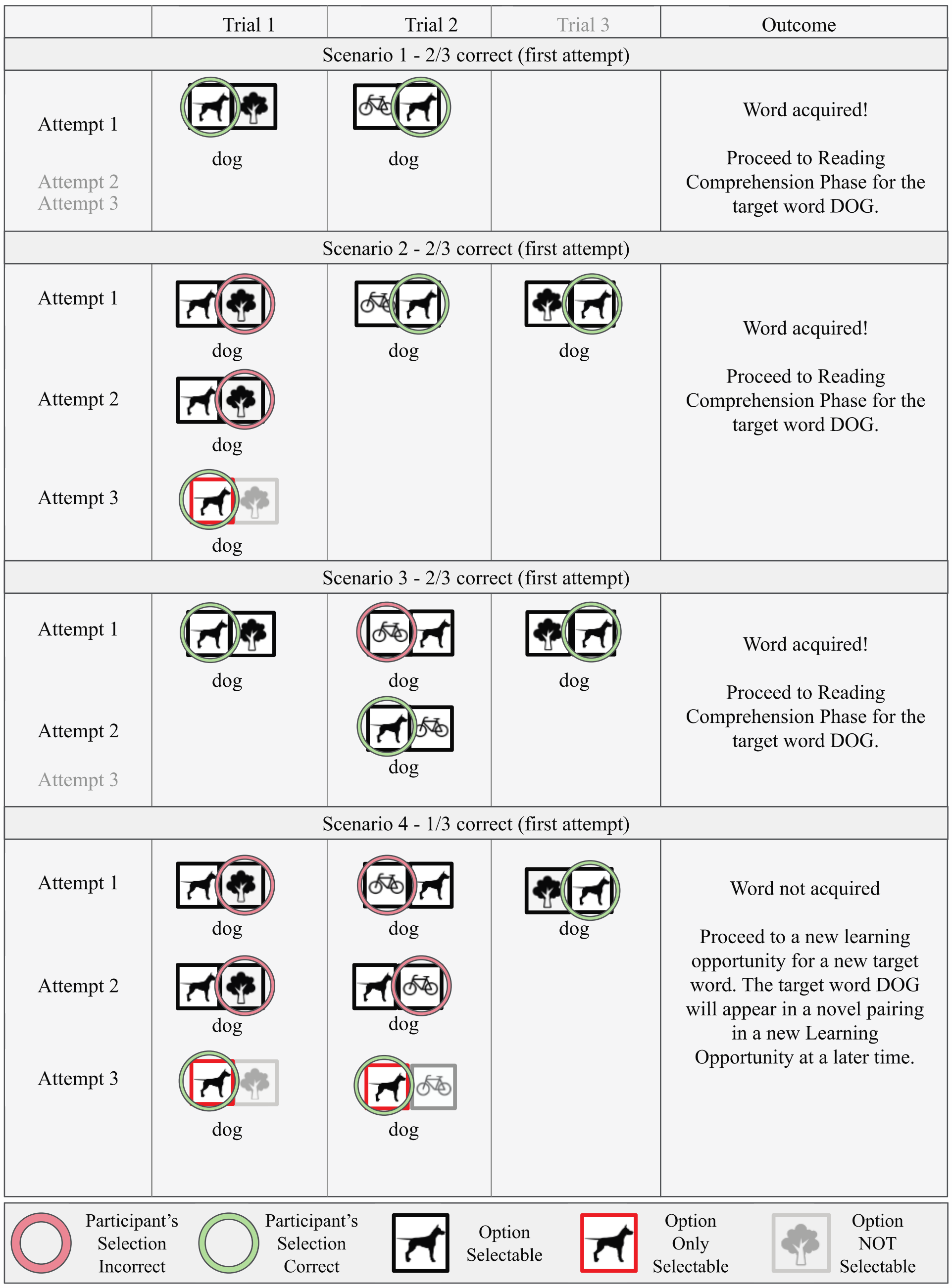


Figure B3

Movement from single word to phrase to sentence level

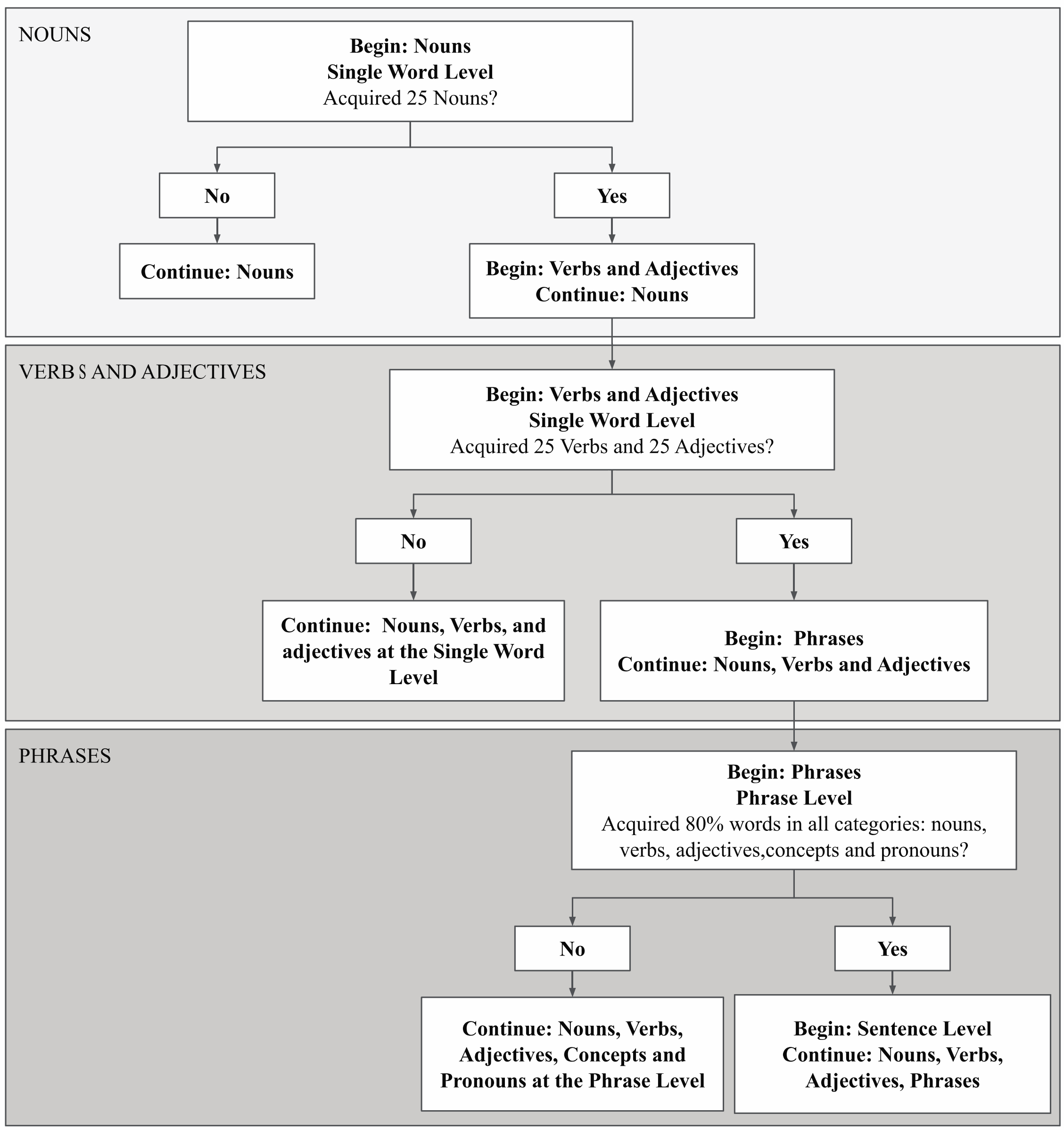


Table B4

Distribution of Participants' Final Level Attained (Word, Phrase, Sentence) for Oral Language and Reading Comprehension by Group

	ASD+HPL*		**ASD-HPL**		**TD**	
Level	Oral	Reading	Oral	Reading	Oral	Reading
Word	0	0	3	3	3	3
Phrase	2	2	2	2	6	6
Sentence	6	6	2	2	6	6
Total	8	8	7	7	15	15

Note: Significant difference* $p<.05$

Chapter 5: Discussion

Emergent Literacy Skills

Objectives and Contributions

For the general preschool population, early literacy development is significantly associated with future language development (Dickinson et al., 2012), social adjustment (Gibson et al., 2020), and academic success (Zimmerman et al., 2008). For preschoolers with ASD+HPL, advancement in these domains may hinge, to some extent, on the ability to leverage their early word reading strength to improve comprehension challenges. Thus, this dissertation has focused on gaining a greater understanding of emergent literacy to better harness early word reading to improve oral and reading comprehension as a means to establishing a path of lifelong learning and achievement.

This investigation into emergent literacy skills in preschoolers with ASD+HPL began with the scoping review in Chapter 2, focused on clarifying cross-study discrepancies and illuminating the early literacy skills of this population. This preliminary scoping review highlighted numerous challenges in studying this population including: small sample sizes; older TD comparison groups; few studies based solely on preschoolers; issues with standardized testing and the language of study, along with discrepant and inconsistent use of terminology across studies. The issue of a lack of a consensual definition for HPL had been discussed in a previous systematic review (Ostrolenk et al., 2017) and this scoping review extended this discussion to additional definitional challenges with nonword reading, phonological awareness and alphabet knowledge that are central to the study of hyperlexia. Results from the literature review also contributed to the debate surrounding the status of phonological awareness and language skills in preschoolers with ASD+HPL and their associated pathway to reading.

Following from the literature review, the primary goal of Article 1 in Chapter 3 was to elucidate the emergent literacy skills and the accompanying reading trajectory of preschoolers with ASD+HPL as either similar to, or distinct from their TD peers, as well as their peers with ASD-HPL. To do so, a number of factors were considered. For example, this was the first study to examine emergent literacy skills in an exclusively preschool sample with numbers greater than one or two participants. Furthermore, the influence of task complexity or task instructions on the ability to demonstrate phonological awareness acquisition could not be ruled out. Thus, a standardized, receptive phonological (rather than phonemic) awareness task, developmentally appropriate for preschoolers with ASD+HPL was used to parse this debate. In addition, correlations between receptive vocabulary and phonological awareness, as well as phonological awareness and reading were examined. The goal was to clarify the discrepancies across previous studies while gaining a deeper understanding of whether vocabulary (reflecting lexico-semantic knowledge) influenced acquisition of phonological awareness (Westerveld et al., 2018; Zhang and Joshi, 2019), or whether phonological awareness skills have not been acquired and are not a necessary condition for word reading (Dynia et al., 2017; Smith Gabig, 2010). This study weighed in on this debate by offering a perspective on language skills and associated correlations for preschoolers with ASD+HPL.

Further, oral language skills of the group with ASD+HPL were examined and compared to both the TD group and the group with ASD-HPL because of discrepancies found across previous studies, but also given the importance of language to reading development (Catts et al., 2006; Chaney, 1992; Ricketts et al., 2013; Snowling et al., 2020). Additional correlations were explored to clarify the relationship between language, preliteracy skills and ASD symptomology. For example, previous research with preschoolers with ASD without HPL had found correlations between language levels and acquisition of phoneme-grapheme correspondence skills (Davidson & Weismer., 2014; Lanter et al., 2012). Thus, Article 1 examined this relationship for preschoolers with ASD+HPL. Article 1 also sought to investigate the relationship between ASD severity and literacy skills. Although previous studies of young children with ASD demonstrated a negative relationship between ASD severity and development of phonological awareness (Davidson & Weismer, 2014; Nally et al., 2018), such that increasing ASD severity was associated with weaker development of phonological awareness, no study, as yet, had looked at this relationship for preschoolers with ASD+HPL.

Article 1 also aimed to clarify the alphabet knowledge profile of preschoolers with ASD+HPL. In doing so, Article 1 contributed to greater awareness of the distinct achievements associated with alphabet knowledge in this population. This served to operationalize alphabet knowledge as separate constructs of phoneme-grapheme correspondence and letter naming in this population, rather than collectively as “alphabet knowledge”. Likewise, this was the case for types of nonwords that were noted to vary considerably across studies. Clarification of these terms ultimately provides more accurate definitions, subsequently leading to more appropriate conclusions drawn from cross-study assessment of these skills. Following from these definitional challenges, Article 1 also contributed to an increased awareness of the challenges inherent in standardized tests that may not represent a valid measure of the target construct, at least for certain populations.

Finally, associations between age, phonological awareness, cognition and reading were explored. Previous correlations had been found between age and the development of phonological awareness for TD children (Anthony & Francis, 2005), as well as age and early word reading for young children with ASD (Nally et al., 2018). By focusing exclusively on a group of preschool children, this study was able to examine this relationship in the absence of the influence of reading instruction. Article 1 also aimed to shed light on the nature of the relationship between IQ and early reading. Although the literature general alludes to a lack of association between IQ and word reading, largely driven by the wide variation in IQ across studies of preschoolers with ASD+HPL with advanced word reading, no previous study had explicitly examined this relationship.

To summarize, the scoping review and Article 1 contributed to the very limited body of existing research on the emergent literacy skills of preschoolers with ASD+HPL. Previous research had studied aspects of early reading in preschoolers with ASD that had not been fully explored for preschoolers with ASD+HPL. Earlier studies had also demonstrated mixed findings in many domains for both ASD populations (ASD+HPL and ASD-HPL). By examining the relationship between language, phonological awareness and phoneme-grapheme correspondence skills; considering the influence of language, ASD severity, age and IQ; clarifying the nature and level of emergent literacy acquisition, and reviewing the cross-study operationalization of terms, together the literature review and Article 1 served to advance current knowledge on emergent literacy for preschoolers with ASD+HPL.

Summary of Results and Intepretation

The initial hypothesis of Article 1 was that preschoolers with ASD+HPL would have weaker phonological awareness and letter-sound correspondence skills as compared to their TD peers and the group with ASD-HPL. The absence of statistically significant differences among the three groups for phonological awareness and letter-sound correspondence was clinically meaningful. The group with ASD+HPL had the lowest mean scaled score on the NEPSY-II for phonological awareness (M=5) of all the groups (ASD-HPL, M=6) and fell below the normative range (8-12) as compared to the TD group (*M*=8). In addition, the group with ASD+HPL had mean scores on letter-sound correspondence as low as the comparison groups. In other words, TD preschoolers who, for the most part, had not yet started reading still had average phonological awareness and emerging letter-sound correspondence skills. In contrast, the preschoolers with ASD+HPL, who exhibited advanced word reading, demonstrated relatively weak phonological awareness and letter-sound correspondence acquisition. This suggests that phonological awareness and letter-sound correspondence have not yet been acquired although word reading is well established. These findings are in agreement with the initial hypothesis of Article 1, as well as those studies of young children with ASD (Dynia et al., 2017; Nally et al., 2018; Smith Gabig, 2010) and preschoolers with ASD+HPL (Cardoso-Martins & Ribeiro da Silva, 2010; Newman et al., 2007) where poor acquisition of phonological awareness has also been found.

However, these findings contrast with those of Westerveld et al. (2017) who found phonological skills in the average range for preschoolers with ASD, as did Newman et al. (2007) for their older participants with ASD+HPL. The older participants in the Newman study may have influenced outcomes, given the impact of literacy instruction on phonological awareness development (Anthony & Francis, 2005). In addition, the Newman et al. (2007) study found the group with ASD+HPL shared a similar language, phonological awareness and cognitive profile to the TD group. Cardoso-Martins and Ribeiro da Silva (2010) questionned whether Newman's hyperlexic group was indeed hyperlexic, raising the issue of varying definitions for the construct of hyperlexia (Ostrolenk et al., 2017). Westerveld et al. (2017) did not have a TD comparison group, therefore, it is difficult to know whether their group with ASD was also more similar to a TD group, which might have explained the finding of typical phonological awareness development in their preschool group with ASD. This speaks to the importance of considering age, and age-matched TD comparison groups, as well as the need to operationalize the term “hyperlexia” when studying this population.

An additional hypothesis of Article 1 was that phonological awareness, alphabet knowledge and oral language skills would not correlate with word reading for the ASD groups as they would for the TD group. In support, the results of the correlation analyses revealed no significant relationships between receptive vocabulary/receptive language and phonological awareness, phoneme-grapheme correspondence skills, nor between phonological awareness, language and word reading for the group with ASD+HPL. These findings suggest that complexity of task instructions is not a factor in demonstrating phonological awareness acquisition, in contrast to Zhang and Joshi (2019). However, their meta-analytic review included studies with older participants with diagnoses other than ASD and comorbidities with ASD that might have influenced outcomes.

Findings from Article 1 also indicated that both receptive and expressive language skills for the group with ASD+HPL were below average and lower than both comparison groups. Although this was partially a consequence of the inclusion criteria applied, Needleman (1982) also described a language impairment in her definition of children with ASD+HPL. Thus, contrary findings of average language abilities in previous studies may be a factor of less stringent identification criteria applied to hyperlexia (Ostrolenk et al., 2017), specifically when the hyperlexic group tends to resemble the TD group (Newman et al., 2007). However, this may also be a function of the heterogenous nature of ASD (Baio et al., 2018) whereby not all have a comorbid language disorder (Carlsson et al., 2013). More studies of preschoolers with ASD+HPL are required to answer this quesiton.

Regarding alphabet knowledge, Article 1 also confirmed that letter naming was significantly stronger for the group with ASD+HPL than the other two groups. These findings were in agreement with previous studies (Cardoso-Martins & Ribeiro da Silva, 2010; Knight et al., 2019; Lamonica et al., 2013; Westerveld et al., 2017) that showed this discrepancy within alphabet knowledge in favour of stronger letter naming over letter-sound correpondence skills. This is particularly relevant for tests which combine both letter naming and letter-sound correspondence under the umbrella category of alphabet knowledge. It also speaks to the nature of metalinguistic abilities in this population. That is, the rote nature and regularity of letter naming (i.e., the letter name is always the same) in contrast with the more abstract, metalinguistic task of letter-sound correspondence (i.e., where the letter name and the associated sound may vary), suggest weaker metalinguistic development.

Finally, cognition is another hetergenous construct found across previous studies of ASD. The mean and range of FSIQ found in preschoolers with ASD+HPL in Article 1 was lower and narrower than had been found in other studies (Grigorenko et al., 2002; Newman et al., 2007), although our inclusion criteria for hyperlexia likely affected this finding. Regardless, FSIQ was shown to be unrelated to word reading in preschoolers with ASD+HPL. In addition, neither age nor ASD symptomology was found to be significantly related to word decoding nor phonological awareness acquisition for preschoolers with ASD+HPL, contrary to findings of Nally et al. (2018) for preschoolers with ASD. However, Article 1 found that the group with ASD-HPL actually had more severe symptomology than the group with ASD+HPL which may have accounted for the diversity across these two studies. Again, more studies are required to explore this relationship, given the heterogeneity across ASD.

Taken together, findings imply that preschoolers with ASD+HPL follow a distinct reading pathway from their TD peers. This distinct approach resembles a more whole-word/orthographic method of reading, rather than a phonological approach taken by TD children. For TD readers, Murray et al. (2019) refer to hierarchical versus sequential-type decoders. Hierarchical decoders make use of spelling patterns and adopt an orthographic approach, whereas sequential decoders read using a letter-by-letter phonics approach. The notion of a hierarchical or pattern-based approach to reading may also apply to early readers with ASD+HPL as per the hyper-systemizing theory (Baron-Cohen, 2006), Enhanced Perceptual Functioning theory (Mottron et al., 2006, 2009) and Veridical Mapping (Mottron et al., 2013). Accordingly, those with lower language and IQ levels may have a higher systemizing mechanism or need to impose patterns as a compensatory mechanism (Baron-Cohen, 2006). In other words, a strong requirement on the part of a young child with ASD+HPL for regularity and consistency may be a function of reduced linguistic and cognitive ability which translates to a pattern-focused approach to word decoding. The significantly lower language and FSIQ levels found in the sample of preschoolers with ASD+HPL in Article 1, as compared to their TD peers would corroborate this theory.

Another possible factor that might explain the underlying poor phonological awareness development is atypical auditory processing of speech stimuli that has been found for children with ASD (O’Connor, 2012). Phonological awareness takes place at an auditory level and speech perception skills have been shown to predict phonological awareness development by the end of kindergarten in 4- and 5-year old children with speech sound disorders (Rvachew, 2006). Similarly, in TD children and children with dyslexia, auditory sensory processing has been linked to the development of phonological awareness (Goswami et al., 2020). Therefore, it is possible that weak auditory processing of speech sounds may account for the challenges seen in the development of phonological awareness for preschoolers with ASD, and lead to a reliance on other strategies or systems to support word decoding. However, comorbid oral language deficits (Carlsson et al., 2013) and attentional challenges (Vivanti et al., 2017) often present in preschoolers with ASD, might confound the task of reliably assessing auditory processing of speech sounds in preschool children with ASD+HPL, rendering this a difficult task for future research.

Reading Comprehension Intervention

Objectives and Contributions

The aim of Article 2 was to evaluate the effects of a novel, tablet-based, parent-supported reading comprehension intervention on oral language (receptive and expressive) and reading comprehension for preschoolers with ASD+HPL. There are currently no early interventions for preschoolers with ASD+HPL that focus on reading comprehension, although a number of computer-based interventions targeting vocabulary instruction for preschoolers with ASD have been reported in reviews of reading comprehension interventions (Khowaja & Salim, 2013). This novel, reading comprehension intervention incorporated early single-word reading using a text-picture matching task, that is believed to be the first successful reading comprehension intervention to begin at the word level for preschoolers with ASD+HPL. This represents a paradigm shift in teaching reading comprehension at a much earlier time period in the preschool years, specifically for educational systems where reading comprehension is typically targeted in the later primary grades (Ministry of Education, Ontario, 2006; Ministère de l'Éducation, Québec, 2002). Thus, by focusing on reading comprehension at the preschool level, Article 2 contributes to those studies advocating for, and demonstrating the benefits of, earlier intervention for children with ASD.

Article 2 also contributes to the larger body of literature concentrated on early intervention as a means to potentially improve long-term outcomes. In view of the documented challenges in reading comprehension (Frith & Snowling, 1983; Ricketts et al., 2013) and social skills (American Psychiatric Association, 2013), alongside correlations between poor social skills, weak academic achievement and behaviour challenges in the classroom (Nadeem et. al. 2010), oral language and reading comprehension intervention to ameliorate and prevent negative social and academic consequences are critical for children with ASD. For TD children, targeting oral language skills in the early years has been found to positively impact narrative skills (a component of social language development) and oral language skills (Fricke et al., 2012), whereby the latter has been shown to support reading comprehension (Catts et al., 2006, 2015). The long-term effects of this short-term intervention cannot be known immediately, however, there is potential for them to extend to social, academic and behavioural domains, and beyond children with exceptionalities to TD children as well.

Article 2 also contributes to the growing body of literature on strength-based, rather than deficit-based, approaches to intervention for children with ASD (Mottron et al., 2009; Remington & Fairnie, 2017). However, hyperlexia is not only a strength, but is also a significant special interest for children with ASD+HPL. As such, intervention targeting early word reading in this group satisfies several criteria that were previously unaddressed. Firstly, young children with ASD+HPL have an intense early interest in letters and words as compared to their TD peers (Ostrolenk, 2018) combined with advanced early word reading skills. It has been noted that this advanced word reading skill plateaus over time until it is on par with TD peers around 10 years of age (Grigorenko et al., 2002). A lack of encouragement or focus on reading has been offered as an explanation for this reduction in skill over time (Grigorenko et al., 2002). Therefore, earlier intervention, administered at the first signs of hyperlexia in the home, may contribute to maintaining a high interest level, and a strong motivation to read, while developing competency in comprehension and prolonging the strength in this domain beyond the primary school years.

Secondly, parents have reported that their children's word reading is not well supported in the early academic years (Newman et al., 2007). Consequently, an early intervention that can be administered successfully by parents at home may contribute to offseting the lack of targeted instruction in the classroom. However, ease of implementation may also contribute to its use by classroom teachers as an adjunct to reading therapy for children with ASD+HPL. That is, using the strength in word reading of young children with ASD+HPL to read to their peers is a constructive practice which is undeniably pleasurable for the child with ASD+HPL. However, for young children with ASD+HPL to actually derive benefit from the practice, reading comprehension should be targeted simultaneously.

Summary of Results and Intepretation

The initial hypothesis of Article 2 stated that preschoolers with ASD+HPL would improve their reading comprehension during a 6-week intervention period (Time 2), when comparing a pre-intervention condition (Time 1-2) to a post-intervention condition (Time 2-3), as compared to the two control groups. The findings supported this hypothesis, demonstrating a significant increase in reading comprehension scores at Time 3 (post-intervention), as measured on the WJ-IV-ACH PC subtest (Schrank et al., 2014; symbol and text representation) for the preschoolers with ASD+HPL, but not for the group with ASD-HPL nor the TD group. Given there was no significant difference across the three groups for reading comprehension at Time 1, this finding was particularly encouraging. It demonstrated that early word-picture matching, to accelerate reading comprehension in preschoolers with ASD+HPL, can be an effective strategy.

Reasons why this intervention might have been successful were explored in Article 2 and included parental support, daily practice and features of the tablet/child interaction. Yet, it is unclear to what extent the success of this intervention is reliant on the tablet format. A recent meta-analysis found that there was no advantage to using iPads to deliver academic interventions in literacy or math, even for male students (Larwin & Aspiranti, 2019). While the findings of Article 2 are a contradiction, future studies may explore the efficacy of alternate forms of delivery of word-picture matching to support oral and reading comprehension. For example, at least one recent study of emergent literacy intervention for preschoolers with ASD found that interactive book reading was instrumental in improving listening comprehension (Hudson et al., 2017). As improvements in receptive language skills for all three groups were significantly higher post-intervention (at Time 3) than pre-intervention at Times 1 and 2, it is possible that the instructions provided to parents to treat the app as they would a book, (i.e., talking about pictures, relating pictures to shared experience) contributed to improved outcomes in listening comprehension. Thus, future studies might investigate physical books or e-books as vehicles for explicit text-picture matching intervention.

Additionally, listening comprehension benefits were found not only for preschoolers with ASD+HPL, but also for the preschoolers with ASD-HPL and their TD peers. It is worth noting that the TD group had quite a wide range of language skills, with some participants scoring in the lower ranges, suggesting that some may have had, as yet, undiagnosed language impairments. Although it was questioned whether receptive language improvement seen for the TD group may have been specific to those with lower language skills, as previous studies have shown that music interventions have a greater impact for children with ASD whose language skills are the weakest (Macdonald et al., 2019), there was no significant correlation between participants with VCI scores lower than 85 and Listening Comprehension scores at Time 3. Nonetheless, this oral language and reading comprehension app contributes to a growing list of easily-implemented interventions advantageous for students both with and without special needs.

Finally, from a research perspective, the significant improvements associated with reading comprehension for the preschoolers with ASD+HPL and receptive language growth for all groups, support both the Simple View of Reading (Hoover & Gough, 1990) and the Dual Coding Theory of Literacy (Sadoski, 2005). Vocabulary development, as a tool to improve reading comprehension (Biemiller, 2003; Carlson et al., 2013), beginning with concrete words with high imagery and supplemented by orthography (Sadoski, 2005) has been shown to be effective for children with ASD. This adds to the growing body of literature (Bell, 1991; Ricketts et al., 2013, 2015) that employs these theoretical models as constructs upon which to base future reading and oral language interventions for children with ASD.

Overall Summary and Contributions

Article 1 clarified the emergent literacy profile of preschoolers with ASD+HPL as compared to their TD peers, and Article 2 demonstrated an effective reading comprehension and receptive language intervention for preschoolers with ASD+HPL. Collectively, they serve to establish effective, recommended teaching practices for this group in home, daycare, clinical and academic settings. This body of work aims to contribute to shaping policy regarding reading comprehension instruction in schools. Understandably, reading comprehension is not a focus of instruction in the early school years when efforts are concentrated on teaching word decoding and phonological awareness (best practice recommended by the National Early Literacy Panel, 2008). Yet, these are practices that do not benefit the child with ASD+HPL. Therefore, the method outlined in these articles provides an easily integrated option to address their area of greatest need.

At a preschool level, this intervention can certainly function as an interim measure for those on a wait list for services; those in rural or remote locations with limited access to services, and those that cannot afford private therapy while awaiting publicly funded treatment. Although children with ASD+HPL represent a small portion of children with ASD (6%-20%, Ostrolenk et al., 2017) and an even smaller portion of the children in a classroom setting (1 in 54; Maenner et al, 2020), the outcomes suggest benefits for young children with ASD and TD children as well. This demonstrates unequivocally why it is vital to pursue studies of populations that are difficult to recruit or may only represent a small subset of the greater population with an exceptionality. As shown, the benefits may extend far beyond their small sample size.

Clinical and Educational Implications

Clinically, at the preschool level, recent trends have targeted earlier evaluation for young children with ASD with the median age range from 29 to 46 months and the initial diagnosis age range of 38 to 57 months (Maenner, et al., 2020). However, the accompanying challenge to earlier diagnosis is increased wait times for treatment. Current wait times for services in Canada and the United States exceed 12 months for children under the age of 5 years old (Brian et al., 2019; Gordon-Lipkin et al., 2016). Growing waitlists in combination with the rising prevalence of ASD (Baio et al., 2018) leave publicly funded agencies scrambling to adjust their intervention models to address lengthy wait lists. It is well documented that the early years are a critical period in development, yet they may be spent on a waitlist. Therefore, an evidence-based intervention that can be easily administered by parents and clinicians, is affordable and meets the needs of preschoolers with ASD, provides an effective stopgap to wait lists as a cost-effective interim solution. In addition, the encouraging findings of increased receptive language scores for the TD group suggest that this intervention may also function as early language support for young TD children as well as those with ASD.

Finally, we anticipate that the significant findings associated with increasing early reading comprehension intervention found in Study 2 will impact recommended teaching practices in the early primary years. As it is well documented that preschoolers with ASD+HPL have reading comprehension challenges, there is little reason to wait until later primary school to address these challenges. In fact, the results of Article 2 suggest that there are significant benefits associated with teaching reading comprehension early, even for 3-year-old children with ASD+HPL.

Limitations and Future Directions

Generalizability of Findings

The primary limitation of our study was the generalizability of our findings given our small sample size for ASD groups. However, as noted previously, recruitment of those with ASD+HPL is challenging (Newman et al., 2007) and studies are typically underpowered (Grigorenko et al., 2002); this was true for Articles 1 and 2 despite recruitment in two separate Canadian provinces. Similarly, the literature review confirmed low sample sizes of single or two participants for previous studies, as did a previous literature review for preschoolers with ASD (Westerveld, 2016). However, there is value in studies with low sample sizes. For example, consider the contribution of the single case study of H.M. (Scoville, 1968) to psychology and the amount of associated research attention this study has generated (Corkin, 1984). Consequently, even studies with small sample sizes can contribute to advancing scientific research and it is encouraging to note that Articles 1 and 2 represent the largest study samples of preschoolers with ASD+HPL to date.

Another factor that affected sample size is the issue of a universally accepted definition of hyperlexia which affects prevalence dependent on the number of criteria used (Ostrolenk et al., 2018). Articles 1 and 2 specifically applied a stringent, five-criteria definition for inclusion in the group with hyperlexia to ensure a valid representation of this population. In doing so, however, this ultimately limited the numbers in the group. That is, more stringent criteria have been associated with lower prevalence (Ostrolenk et al., 2018). Yet, as a result of such stringent criteria, these two studies confidently represent preschool children with hyperlexia in spite of the well-documented heterogeneity in ASD (Baio et al., 2018). As noted, accurate representation of the sample as hyperlexic has been questioned by previous researchers (Cardoso-Martins & Ribeiro da Silva, 2010) in studies with larger sample sizes (Newman et al., 2007). For these reasons, it is recommended that future studies of children with ASD+HPL also adopt Needleman’s 5-criteria for HPL inclusion.

Standardized Assessment Measures and Preschoolers with ASD+HPL

Issues surrounded nonword reading and the standardized measurement used (WJ-IV ACH Word Attack subtest). Firstly, this test assesses both phoneme-grapheme correspondence skills as well as nonword reading. As the focus was to assess both skills while minimizing testing for the very young participants, this measure was adopted. Yet, this meant that only six items for phoneme-grapheme correspondence were available. It would have been preferable to have a larger number of phoneme-grapheme correspondence items to sample from. In addition, some participants with ASD+HPL obtained a ceiling (of six incorrect), likely due to their challenges with phoneme-grapheme correspondence skills, making it unclear whether they could have gone on to read nonwords. In retrospect, administration of the test should have been adapted to allow participants to do both phoneme-grapheme correspondence and nonword reading (without imposing the ceiling). As only three participants in our group with ASD+HPL were able to read nonwords, we were unable to draw conclusions about this skill. Another option would have been to use a separate standardized nonword reading assessment to enable a more thorough exam of this domain and permit analysis of the type of words that preschoolers with ASD+HPL were able to read. It would be beneficial to examine word type in future studies.

More specifically, based on previous findings of intact nonword reading for preschoolers with ASD+HPL in the presence of challenges with the methodology of assessing this construct (i.e., orthographic depth of the language, type of nonwords assessed, regular vs irregular words) highlighted in the literature review, Articles 1 and 2 did not enrich the dialogue in this domain. Furthermore, our discussion of the hyper-systemizing theory (Baron-Cohen, 2006) alongside the Enhanced Perceptual Functioning theory (Mottron et al., 2006, 2009) to explain an alternative, pattern-based approach to reading in preschoolers with ASD+HPL could have been supported or refuted based on a more in-depth analysis in this domain. For example, if regular nonwords, (i.e., those with a regular word shape) were found to be easier to read than irregular nonwords (or those with an irregular pattern), this would lend support to a pattern-based approach. It is hoped that future studies will delve deeper into this area with preschoolers.

Higher-order Cognitive Functions and Word Reading

Although it was determined that there was no correlation between FSIQ and word reading for preschoolers with ASD, it was not determined if this was also the case for NVIQ and reading. Previous studies with preschoolers with ASD have shown that NVIQ predicts word reading (Davidson et al., 2014; Westerveld et al., 2018). The particular challenge for Article 1 revolved around the WPPSI-IV^{CDN} subtests to determine composite domains. That is, the subtests administered to determine FSIQ were also the same ones that determined NVIQ with the 3- to 4-year old children, but not with the older group. This meant that an extra subtest would have had to be administered to a portion of the group to obtain NVIQ for all participants. Cognizant of the limitations of the very young participants to engage in standardized testing, the decision was made to not administer an extra subtest and thus the opportunity to conduct this analysis was waived. However, when scores from the Block Design subtest of the WPPSI-IV^{CDN} are used as a proxy for NVIQ, there are no significant correlations between NVIQ and word reading for the group with ASD+HPL (r=.135, p=.583), but the group with ASD-HPL did show a significant negative correlation, r=-.874, p=.023, suggesting higher word reading scores are associated with lower scores on the Block Design subtest. Based on these preliminary findings, it is hoped that future studies will address this question of the association between NVIQ and word reading in preschoolers with ASD+HPL and ASD-HPL.

Intervention Challenges

The novel reading comprehension intervention app was designed and built to capitalize on the special interests of the population with ASD+HPL. As these children's interest in reading is already high, parents reported no challenges in soliciting their participation on a daily basis. However, this was not the case for the TD participants, as well as some participants with ASD-HPL. This was likely due to multiple factors. Firstly, less than half the TD children (30%) and those with ASD-HPL (42%) were reading yet, and so were less successful and consequently, less motivated by a reading app. Second, there were very few "bells and whistles" built into this app as potential distractions. Previous research has shown that certain features of an app in e-books can distract from the task, particularly with language-impaired children, and lead to an increase in management-type dialogue on the part of parents (Rees et al., 2017) as compared to preferred narrative-type dialogue that's focuses on the content of the story. To avoid this scenario, the app was developed to minimize distracting features. In doing so, however, the TD children and those with ASD-HPL, were potentially less motivated to participate, especially with lower reading skills and less interest in letters and words than their peers with ASD+HPL. Therefore, this may have affected results for these groups.

Furthermore, as noted in Macdonald et al. (2020) [Article 1] children with ASD+HPL are likely not reading via a phonological approach but rather using a more sight word technique. The DCT is aligned with a sight word reading approach and comprehension was potentially facilitated by this focus. However, this was likely not the case for the TD children, and it is unclear which approach is favoured by the group with ASD-HPL. The whole word/orthographic reading approach adopted in this app was not best practice for teaching word reading to TD children who benefit from a phonological or phonics-based reading approach (Brady, 2011). Therefore, their TD child and the child with ASD-HPL's potential frustration in combination with their children's preference for other activities associated with the digital device, may have contributed to poorer outcomes for the TD group and the group with ASD-HPL in reading comprehension. This app is likely more appropriate for the TD group and the group with ASD-HPL once they have begun to read and might support reading comprehension in children who do not respond to a phonological approach to reading. However, findings of improved scores for receptive language skills indicate that despite any lack of engagement, there were still benefits to the TD group as well as the group with ASD-HPL. Future studies should adapt and explore this intervention with TD children and those with ASD-HPL who are reading, or with young children with a reading impairment in comprehension or decoding.

Concluding Remarks

The literature review, and both Article 1 and 2, represent a body of work that advances our knowledge of these remarkable children with ASD+HPL. Not only is their reading profile clarified by this work, but a recommended teaching practice is now available that supports their reading comprehension and language challenges, while capitalizing on their areas of strength and promoting their intense interest. In addition, this intervention is successful at the very early preschool stage when implemented by parents, which bodes well for early intervention and later academic and social success. Finally, early improvement in reading comprehension offer children with ASD+HPL the opportunity to benefit from reading, not only for the pleasure of decoding words, but at a deeper, more meaningful and enriching level that opens the door to learning.

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