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Reading Comprehension Development in Children with Intellectual Disabilities

Evelien van Wingerden - Fontein

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Reading comprehension in children with intellectual disabilities

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1.

General Introduction

Learning to read is essential for anyone who wants to participate in daily life. Being able to read enables autonomy and enhances self confidence as it enables learning and information gathering, gives a broader world view, and – for children - reduces the dependence on parents or caregivers to provide information. For individuals with a mild intellectual disability (ID), however, being able to read and comprehend written text is more challenging than for typically developing children. It is estimated that around 1% of all children has an ID, of which 64% are categorized as mild ID (Van Naarden Braun, et al., 2015). Children with mild ID are able to gain academic skills, but experience significant difficulties in processes such as in planning, problem solving and memorizing information. Therefore, they have more difficulty in achieving higher-order skills such as reading comprehension. A complicating factor in literacy attainment is the high rate of additional disabilities in individuals with ID as part of their genetic condition (e.g., Down syndrome) or because of additional conditions (autism, cerebral palsy, sensory impairments). The prevalence of moderate hearing loss is 3.0 % in children with ID compared to 0.14% in children with average intelligence (Van Naarden Braun, et al., 2015). The reading development of children with ID has not yet been documented for a large group of children with varying etiologies, and from a longitudinal perspective. In order to provide an outline for optimal reading education, it is necessary to gain insight in the reading process of children with mild ID and their main pitfalls in reading comprehension. The present thesis will explore the precursors for reading skill and reading comprehension in children with ID, and in children with ID who are Deaf or Hard of Hearing (DHH), referred to as children with ID-DHH in this thesis. The thesis includes cross-sectional and a longitudinal analyses to identify the main cognitive and linguistic precursors for reading comprehension in children with mild ID.

Typical reading comprehension

To comprehend a text means to construct a meaningful text representation in perspective of a certain reading goal. The most comprehensive framework of the reading process to date is the reading systems framework (Perfetti, 1999; Perfetti & Stafura, 2014). This framework incorporates several previous models of reading, including the simple view of reading (Hoover & Gough, 1990). One main characteristic of the reading systems framework is a distinction between word identification and comprehension processes. The simple view of reading states that reading comprehension requires two types of skills: being able to translate written words to their spoken equivalent (decoding), and to comprehend the meaning of the text (listening comprehension). Word decoding is typically described as matching letters to corresponding speech sounds, and blending the letter sequences

to match phonological words. This requires letter knowledge, understanding of sounds within spoken language (phonological awareness), rapid naming skills, and verbal short-term memory (Melby-Lervåg, Lyster, & Hulme, 2012; Moll, et al., 2014). Listening comprehension can be defined as matching words to their plausible meanings, and integrating these concepts into a meaningful message. This requires a robust vocabulary, grammar comprehension, reasoning skills and verbal working memory (Vellutino, Tunmer, Jaccard, & Chen, 2007; Stevenson, Bergwerff, Heiser, & Resing, 2014). The simple view of reading implies that deficits in one of the two domains will suppress the level of reading comprehension, but also that stronger skills in one domain can compensate for weaknesses in the other domain.

Word decoding and listening comprehension are considered independently developing skills in early readers. While language comprehension will evolve naturally for most persons growing up in a language-rich cultural context, decoding is an abstract skill and needs to be explicitly learned by all readers. As described in the convergent skills model of reading development, the relative contribution of different underlying skills and abilities to reading comprehension changes over time (Vellutino, et al., 2007). In the earlier stages of learning to read, the emphasis lies on learning to decode. The development of letter knowledge and phonological awareness, which are referred to as 'early literacy skills' in the present thesis, are partly learned before the start of formal education, through direct and indirect literacy experiences at home (Sénéchal & LeFevre, 2002). The start of formal reading education is focused on further internalizing the letter-sound correspondences and the process of blending phonemes into words. In more skilled readers word reading does not require explicit decoding, but can be achieved through direct, orthographic recognition (Seidenberg & McClelland, 1989). Once word decoding skill is mastered and requires less cognitive resources, reading comprehension becomes more dependent on the level of listening comprehension (e.g., Landi, Frost, Menc, Sandak, & Pugh, 2013; Verhoeven & Van Leeuwe, 2008).

Decoding and listening comprehension are connected through the lexicon: knowledge of the meaning, morphology and syntax of words. The lexical quality hypothesis (Perfetti & Hart, 2002) emphasizes that reading comprehension is related to decoding and language comprehension, which are both connected through vocabulary (see also Protopapas, Mouzaki, Sideridis, Kotsolakou, & Simos, 2013; Language and Reading Research Consortium [LRRC], 2015). Within the lexical quality hypothesis, vocabulary has a reciprocal relationship with both word recognition and comprehension (Perfetti, 2007, 2010). The theory states that better in-depth knowledge about the words in the mental lexicon will improve reading and comprehension of text meaning. This includes semantic information, but also orthographic and phonological knowledge. More knowledge about a word will result in well-specified representations of orthography and phonology, alongside diverse and flexible representations of meaning. Word representations of high lexical quality will improve word decoding and word recognition, and text comprehension. Lower lexical quality representations will lead to word-related problems in reading and comprehension. Reversely, a reader with good comprehension skills can also learn new word meanings and expand their vocabulary while reading. Decoding skill strengthens the connection between wordform and meaning, and also expands vocabulary by providing a context of surrounding words.

Finally, the construction-integration model (Kintsch & Van Dijk, 1978) describes three levels of reading comprehension. The first level of comprehension is the construction of a semantic representation of the text, based on the literal meaning of individual words and phrases. At the second level, words are combined into propositions and sentences, which can then be connected into a coherent text representation (word-to-text-integration). If the text contains implicit or ambiguous information, the reader draws inferences about the meaning of words and sentences in the given context, using semantic and general knowledge. At the highest level of comprehension, the text representation progresses into a situation model, a mental model that relates the contents and meaning of the story to the reader's prior knowledge, goals and memories. (Hannon & Daneman, 2004; Kintsch & Van Dijk, 1978).

Throughout the reading process the reader relies on existing knowledge sources: orthographic knowledge, linguistic knowledge and general knowledge. Retrieval and integration of this knowledge takes place within a cognitive system of memory pathways that has limited processing capacity (Perfetti & Stafura, 2014). In other words, a person's knowledge base and cognitive abilities such as long-term and short-term memory, attention, processing speed, and reasoning ability are prerequisites for successful reading comprehension. Restrictions in the cognitive system can also influence the processing of sensory input and the development of the knowledge base itself. For example, temporal processing skill facilitates the expansion of phonological knowledge through its role in speech perception (Huss, Verneij, Fokster, Mead, & Goswami, 2011) and is also related to language comprehension through its role in detecting prosody in spoken language (Gordon, Jacobs, Schuele, & McAuley, 2015). Also, word decoding is related to processing speed and integration of linguistic and perceptual reading-related processes, as is reflected in rapid naming tasks (Norton & Wolf, 2012).

The comprehension of explicit text content relies heavily on orthographic and linguistic knowledge as well as foundational cognitive skills such as working memory, pattern recognition, temporal processing and attention (Malenfant, Grondin, Boivin, Forget-Dubois, Robaey, & Dionne, 2012; Stevenson, Bergwerff, Heiser & Resing, 2014). A deeper understanding of a text demands more knowledge resources and cognitive skills; on top of strong linguistic knowledge a good situation model requires additional resources such as a larger general knowledge base and higher-level cognitive skills, for example inference making, executive functioning and attention allocation (Fuchs, Compton, Fuchs, Bryant, Hamlett, & Lambert, 2012; Kendeou, Van den Broek, Helder, & Karlsson, 2014; Kim, 2016; Sesma, Mahone, Levine, Eason, & Cutting, 2009). Many of these are gained through implicit learning; by observing other persons who communicate. To extract these conventions correctly, requires reasoning ability, working memory capacity, and attention (Kendeou, et al., 2014).

Reading comprehension in children with an intellectual disability

The present research focuses on children with mild ID. Individuals with mild ID are defined as having deficits in adaptive behavior, but are able to achieve comparative independence in daily life and gain academic skills at or above an elementary level, provided that they receive sufficient support (American Psychiatric Association, 2013). The IQ of individuals with mild ID generally lies between 50 – 70, however in practice there is a group of individuals who were previously defined as 'borderline ID' (IQ between 70-80; American Psychiatric Association, 2000) who have additional conditions that result in a similar level of cognitive and adaptive functioning as individuals with mild ID (Woittiez, et al., 2014). The reading level is highly variable among children with mild ID (Lemons, et al., 2013). This variation is partly linked to the severity of ID (Levy, 2011), but individual variation is largely unexplained. To provide optimal support in reading education, it is necessary to know the main precursor skills for reading in children with mild ID. Several studies have investigated subskills of reading comprehension (mainly word decoding; Chanell, Loveall, & Conners, 2013; Soltani & Roslan, 2013) and vocabulary; or studied sub-populations within the general ID population (Antschel, Hier, Fremont, Faraone, & Kates, 2014; Nash & Heath, 2011; Roch, Florit, & Levorato, 2011). Many studies have focused on the effectiveness of reading intervention programs (Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2014; Browder, Hudson, & Wood, 2013). However, little is known regarding the fundamental prerequisites of reading development in children with mild ID.

Relating to the reading systems framework (Perfetti, 1999), it can be expected that limitations in cognition may impact the reading process of children with mild ID in several ways. Firstly, their learning disability will restrict the scope of their knowledge sources (in particular general knowledge and linguistic knowledge; Alloway, 2009). Secondly, limitations in cognitive capacity are likely to affect the processing of the text; the interactive use of knowledge sources during construction of the text representation and the drawing of inferences during higher-level text processing. For example, both problem solving ability and working memory updating are weaker in children with mild or moderate ID than in control groups matched on nonverbal IQ (Carretti, Belacchi, & Cornoldi, 2010; Goharpey, Crewter, & Crewter, 2013), which has consequences for the construction of meaningful text representations. This is supported by Numminnen, Service, and Ruoppila (2002) who found that adults with mild ID depended more on knowledge support from long-term memory than typically developing children in a mental age control group, who could benefit more from efficient online working memory processes.

Regarding the general process of reading comprehension, the available information about its precursors in children with mild ID is fragmented. As in typical readers, early literacy skills are crucial for achievements in decoding and reading comprehension (Dessemontet & De Chambrier, 2015). In the preliteracy stage, children with mild and moderate ID are associated with poor phonological awareness (Channell, et al., 2013). At the same time, phonological awareness is a major predictor for word decoding, next to verbal short-term memory (Channell, et al., 2013; Conners, Atwell, Rosenquist, & Sligh, 2001; Soltani & Roslan, 2013) pointing to linguistic knowledge and cognitive capacity as weaknesses in the decoding process for children with mild ID. Remarkably, relationships between vocabulary and decoding have rarely been detected in children with ID (e.g., Dessemontet & De Chambrier), but vocabulary is related to language comprehension (Levorato, Roch, & Beltrame, 2009; Verhoeven & Vermeer, 2006). The vocabulary of individuals with ID increases with chronological age and is generally larger compared with typically developing children of the same mental age (Facon & Bollengier, 2009). The development of higher language processing is more problematic (Facon, Facon-Bollengier, & Grubar, 2002; Levorato, et al., 2009). The notion that cognitive deficits are the reason for language processing difficulties is supported by earlier findings that individuals with ID perform lower when sentences have a complex syntactic structure (Jones, Long, & Finlay, 2006; Nation, 1999) or require inference making (Ezell & Goldstein, 1991; Tavares, Fajardo, Ávila, Salmerón, & Ferrer, 2015). For young adults with Down syndrome, it has also been found that inferential reading comprehension was problematic in comparison to literal comprehension

(Nash & Heath, 2011). One study so far focused on cognitive predictors for a reading comprehension in adolescents with borderline intellectual functioning. It was found that reading comprehension was predicted by decoding, attention, self-monitoring, and working memory (Antschel, et al., 2014). In short, cognitive deficits in individual with mild ID have been shown to affect reading comprehension at the base, through phonological awareness and working memory, and at the higher level, by hampering inference making and construction of a text representation. Less clear are the interrelations between precursors and their relative influence on reading comprehension in children with ID.

Reading comprehension with an ID and an additional hearing loss

A child is considered hearing impaired when hearing loss greater than 30dB in the better hearing ear and deaf when hearing loss is > 81 dB in the better ear. In high-income countries, the prevalence of hearing loss in children is around 0.5% (World Health Organization [WHO], 2012) and the prevalence of hearing loss in children with ID is estimated around 3.0% (Van Naarden Braun et al., 2015). For adults, the prevalence of disabling hearing loss lies around 4.9% in high income countries (WHO, 2012) while the prevalence of hearing loss among adults with mild to moderate ID is estimated from 21% up to 51%, depending on the type of measure and criteria (Carvill, 2001; Evenhuis, Theunissen, Denkters Verschuure, & Kemme, 2001). The prevalence increases with age and is much higher in individuals with Down syndrome (Meuwese-Jongejeugd et al., 2006).

Even when a cochlear implant or hearing aid permits a certain level of hearing, the perception of speech is deficient in children who are DHH (Mayer, 2007). When phonemes cannot be discerned properly in spoken language, word identification by means of grapheme-phoneme mapping is no longer feasible for early readers (Mayberry, Del Giudice, & Lieberman, 2011). Instead, word meanings are used as an anchor to connect signs and orthographic units (Hermans, Knoors, Ormel, & Verhoeven, 2008). Although acquisition of word identification skill is challenging for children who are DHH, it is thought that their main concern in reading comprehension is the process of language comprehension. The language structure of sign language is fundamentally different from the oral language, both in sentence structure and phonological characteristics. Moreover, exposure to sign language or speech for children who are DHH is less continuous than exposure to spoken language for hearing children. This will affect general language development and lexical quality (Coppens et al., 2011; 2012). Wauters, Van Bon, and Tellings (2006) found that reading comprehension levels for children who are DHH are generally below the level of word decoding skills. Reversely, strong language skills in either sign or oral language can provide an anchor for learning to understand written text (Cummins, 1981; Mayer, 2007; Nelson & Crumpton, 2015). Besides the comprehension of grammar and language structure, weak oral language comprehension also hinders the expansion of vocabulary. It particularly affects learning abstract words that need to be learned through language (Perfetti, 2002; Wauters, et al., 2006).

In terms of the reading systems framework, children who are DHH experience phonology-related deficiencies in the orthographic and linguistic knowledge systems. Therefore, the demands increase on the remaining knowledge source (general knowledge) and on the cognitive system in order to reach an appropriate level of reading comprehension. Also, lexical quality is key to reading comprehension for this group; lexical quality is necessary to initiate word recognition and is a major source of support in text interpretation.

For children with ID-DHH, limited access to oral language combined with limited cognitive abilities will lead to problems in all aspects of reading comprehension (knowledge sources, processes of reading, cognitive resources). In addition, in the case of multiple disabilities a deficit in one domain is likely to hinder coping mechanisms to compensate for losses in the other domain. For example, children with ID experience difficulties in interpreting the acoustic signals that are provided by hearing technology through limitations in cognitive capacity and linguistic knowledge. Studies have shown that the effects of cochlear implants on auditory perception and language development are significant in children with mild ID, but much lower in children with moderate or severe ID (Daneshi & Hassanzadeh, 2007; Edwards, 2007). Reversely, limited access to auditory information due to a hearing impairment will restrict the increase of linguistic knowledge, which is already weak in children with ID. These and similar issues lead to 'diagnostic overshadowing' (Carvill, 2001) and make it difficult to distinguish between the effects of ID versus hearing loss on literacy development. This also raises questions about the relative influence of both disabilities on the process of reading in children who are ID-DHH.

The current thesis

Despite the great difficulties that children with mild ID experience in learning to read, written text can have advantages over spoken communication. For example, text is a more permanent form of communication, messages are less volatile than in speech. This provides time to process the text and opportunity to re-read if necessary. If

children with mild ID are able to read age-appropriate texts (adapted if necessary), it expands their vocabulary and world knowledge, and increases independence. It is therefore essential to provide optimal support for reading development. In order to make the greatest progress, they will need opportunities, encouragement and instruction that is suitable to their needs (Erickson, 2005; Kliewer, 2008; Browder, Gibbs, Ahlgrim-Delzell, Courtade, Mraz, & Flowers, 2008).

The exact nature of reading comprehension problems has not been established for children with mild ID. The reading systems framework (Perfetti, 1999; Perfetti & Stafura, 2014), similar to other models of decoding and reading comprehension, does not specify the cognitive components that are involved in the reading comprehension processes. The general learning difficulty may restrict reading acquisition through limited linguistic knowledge and information processing, mainly affecting the level of decoding. At the same time, difficulties in general knowledge, working memory and abstract reasoning may affect top-down reasoning during the building of a coherent story representation, which impairs listening comprehension. In short, a cognitive deficit affects both decoding and listening comprehension (Hoover & Gough, 1990). For children with ID-DHH these disadvantages may very well be enhanced, because hearing problems also harm their spoken language development.

At present, teachers struggle to provide suitable support for the reading development of children with ID. In typical reading education in The Netherlands, children start with decoding orthographically transparent words. This is expanded by word recognition of more opaque words. Once word decoding is sufficiently developed, which is in second grade, reading comprehension (focusing on reading strategies) is introduced. The approach to literacy learning for children with ID is not clearly defined. Some schools use an adapted version of regular educational methods, while others start with symbol recognition and word-recognition, then building on this orthographic knowledge. Unfortunately, the step from orthographic recognition to phonological understanding is difficult to make for many students with mild ID. In practice, much attention goes to word decoding or word recognition, while reading comprehension generally receives less attention as long as word reading skills are still at a lower level. There is little instruction in reading strategies, and reading comprehension is mostly stimulated by independent book reading (Van der Laan, 2006). For children with ID-DHH, no clear guidelines exist for reading comprehension. Education is in many respects similar to education for children with ID, with support from visual aids such as visual symbols and the use of Sign-Supported Dutch. The children learn to read through symbol recognition, and transfer to letter learning and word recognition quite late, when teachers estimate that they are ready to comprehend the concept of an orthographic representation of language.

The research in the current thesis aimed to gain insight in the level of reading comprehension in children with mild ID, and its main cognitive and linguistic precursors. The focus of the research was on the two main processes of reading comprehension, decoding and listening comprehension, which are highlighted in the simple view of reading (Hoover & Gough, 1990). The goal was to mark any specific characteristics in their pattern of reading development and gain insight in their requirements for reading education. Cross-sectional and longitudinal analyses were performed to investigate the relationship between reading comprehension and its main precursors at different levels.

The present thesis aimed to answer the following research questions:

a) How do cognitive and linguistic skills contribute to explicit and implicit reading comprehension performance in children with mild ID?

b) What are the precursors for reading comprehension in children with mild ID and how does their reading profile deviate from children with typical development?

c) What are specific characteristics of reading comprehension in children with ID-DHH and what implications does their hearing impairment have for reading education?

Outline of this thesis

The current research describes four studies that address the research questions above. The reading systems framework (Perfetti, 1999; Perfetti & Stafura, 2014) will be the overarching theoretical framework, with a focus on word decoding and listening comprehension, in line with the simple view of reading (Hoover & Gough, 1990).

Chapter 2 looks at the basic model of reading comprehension of children with mild ID by including word decoding, language comprehension, vocabulary and cognition as predictors for higher-level and lower-level reading comprehension in children with mild ID who have elementary word decoding skills. Their performance on these tests are compared to a control group of typically developing children who had a word decoding level within the same range.

Chapter 3 and 4 take a closer look at the reading comprehension process, using the simple view of reading (Hoover & Gough, 1990) as a starting point. It is tested to what extent the variation in reading comprehension can be explained from word decoding, listening comprehension and precursor measures. Cognitive (rapid naming, phonological short-term memory, working memory, temporal processing, nonverbal reasoning) and linguistic predictors (early literacy skills, vocabulary, grammar) are included. Also, children with mild ID of all reading levels are included in these studies. A cross-sectional and longitudinal study are conducted to explore the main predictors for reading comprehension in children with mild ID. Using structural equation modeling, the relationships between precursor measures and outcome measures are investigated. The resulting reading model is compared to typical predictors of reading comprehension, in order to identify any deviations.

The impact of additional sensory disabilities on reading is explored in Chapter 5. The goal of this study is to understand the impact of hearing impairment in addition to intellectual disability on the reading comprehension development. Children with ID-DHH are assessed on tests that are highly similar to the tests in Chapter 3 and 5, in order to compare their performance to children with normal hearing. Stronger and weaker performers at several levels of literacy are compared to gain insight in the characteristics of stronger readers with ID-DHH. The pattern of their scores is interpreted in the light of existing theories about reading in children with mild ID and in children who are DHH.

Finally, a general discussion and conclusion are provided in Chapter 6. This chapter reviews and discusses the outcomes of the studies in the light of current theories on reading comprehension. In addition, the limitations of the present research are discussed, along with suggestions to improve further research in this field. To conclude, the practical implications of the present findings are presented.

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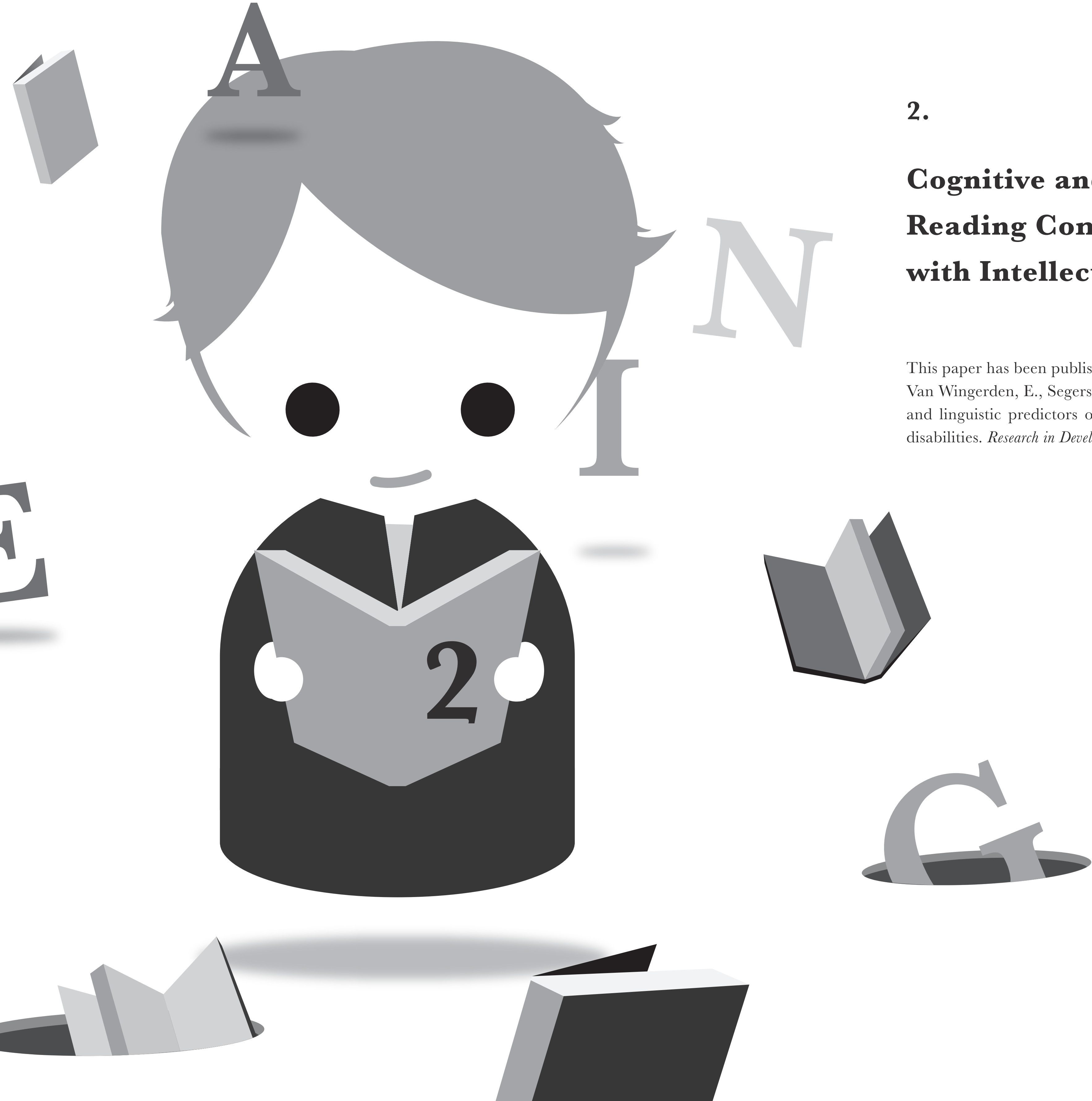

2.

Cognitive and Linguistic Predictors of Reading Comprehension in Children with Intellectual Disabilities

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Abstract

A considerable number of children with intellectual disabilities (ID) are able to acquire basic word reading skills. However, not much is known about their achievements in more advanced reading comprehension skills. In the present study, a group of 49 children with ID and a control group of 21 typically developing children with word decoding skills in the normal ranges of first grade were compared in lower level (explicit meaning) and higher level (implicit meaning) reading comprehension abilities. Moreover, in the group of children with ID it was examined to what extent their levels of lower level and higher level reading comprehension could be predicted from their linguistic skills (word decoding, vocabulary, language comprehension) and cognitive skill (nonverbal reasoning). It was found that children with ID were weaker than typically developing children in higher level reading comprehension but not in lower level reading comprehension. Children with ID also performed below the control group on nonverbal reasoning and language comprehension. After controlling for nonverbal reasoning, linguistic skills predicted lower level reading comprehension but not higher level reading comprehension. It can be concluded that children with ID who have basic decoding skills do reasonably well on lower level reading comprehension but continue to have problems with higher level reading comprehension.

Being able to read is crucial for a person's independence in life, and expands opportunities for gaining knowledge (Boudreau, 2002; Verhoeven, 1994). This also applies to children with Intellectual Disabilities (ID). Despite developmental disabilities, many children with ID are able to acquire basic literacy skills, although their level of literacy can vary widely (Jones, Long, & Finlay, 2006; Kliewer, 2008; Koppenhaver & Erickson, 2003). But being able to read or recognize words does not guarantee comprehension of what is read.

The current knowledge regarding reading comprehension of children with ID is limited. Most research in this area concerned intervention studies, focusing on a wide range of reading comprehension strategies, rather than identifying the underlying reasons for difficulties in reading comprehension (Browder, Wakeman, Spooner, Ahlgrim-Delzell, & Algozzine, 2006; Mason, 2013; Van den Bos, Nakken, Nicolay, & Van Houten, 2007). Also, no clear distinction has yet been made between different levels of reading comprehension in individuals with ID. In order to optimize future intervention programs for children with ID, it is necessary to increase explanatory power and gain more insights regarding specific characteristics of their reading profile. In the present study, we investigated predictors for reading comprehension once word decoding skills were attained. The aim was to distinguish the main predictors for lower level (explicit) and higher level (implicit) reading comprehension in children with ID who were already at first-grade word decoding level.

Typical reading comprehension

Comprehending a text involves connecting the individual elements within the text in order to construct a meaningful message (Kintsch & Rawson, 2005; Oakhill & Cain, 2012). Based on lexical knowledge, the single word meanings can be organized into meaningful propositions. Next, these propositions are connected by using cues within the text, like anaphoric pronouns and adverbs. This allows detection of the underlying text structure and the overall meaning of the text. In addition to comprehension of explicit text meaning (lower level comprehension), full understanding of a text requires reasoning, induction, deduction and resolving of anaphoric ambiguities. A readers needs to draw inferences about the implicit meaning of words and sentences in the context of a particular passage (higher level comprehension; Hannon & Daneman, 2004; Kintsch & Van Dijk, 1978).

One of the leading theories on reading comprehension in the typical population is the simple view of reading (Gough & Tunmer, 1986; modified by Hoover & Gough, 1990), stating that successful reading comprehension is a product of decoding skill and language comprehension. According to the convergent skills model, decoding is the main contributor to reading comprehension during the early stages of literacy, because most cognitive resources are involved in interpreting the graphic symbols. Over time, when decoding becomes an automatic process, language comprehension becomes the main determinant of reading comprehension (Vellutino, Tunmer, Jaccard, & Chen, 2007; Verhoeven & Van Leeuwe, 2008).

Complementary to the simple view of reading, the lexical quality hypothesis states that the degree of comprehension is influenced by the size of the vocabulary, as well as the quality and flexibility of individual lexical representations (Perfetti, 2007; Perfetti & Hart, 2002; Tannenbaum, Torgesen, & Wagner, 2006). Vocabulary indeed is a strong predictor for reading comprehension, even after decoding skill and language comprehension have been controlled for (Cain, Oakhill, & Lemmon, 2005; Ouellette & Beers, 2010; Protopapas, Mouzaki, Sideridis, Kotsolakou, & Simos, 2013).

Recently, several studies have attempted to connect both theories. In a longitudinal study, Verhoeven and Van Leeuwe (2008) observed that in higher grades in Dutch primary schools, only vocabulary directly predicted reading comprehension, while language comprehension influenced reading comprehension through a reciprocal relationship with vocabulary. For Greek, Protopapas et al. (2013) found similar results in a cross-sectional longitudinal study that vocabulary was a strong predictor for reading comprehension in Grade 3-6. In addition, they demonstrated that the predictive value of decoding for reading comprehension may be largely interrelated to language comprehension. These recent studies confirm the importance of decoding skill, language comprehension, and vocabulary for reading comprehension (Hoover & Gough, 1990; Perfetti, 2007). Furthermore, they indicate that reading comprehension is largely attained through a combined contribution of these three linguistic skills.

Reading comprehension is not predicted by linguistic skills only. In addition, a certain degree of cognitive development is required for attaining higher level reading comprehension (Oakhill & Cain, 2012). Reasoning ability has been shown to be a steady, unique predictor of reading comprehension next to linguistic predictors in lower grades of primary school (Fuchs et al., 2012). Cognitive skills enable the reader to relate the linguistic information from the text to their existing world knowledge, monitor text comprehension, and adjust their interpretation if necessary (Hagoort & Van Berkum, 2007; Oakhill & Cain, 2012; Sesma, Mahone, Levine, Eason, & Cutting, 2009).

Reading Comprehension in Children with ID

Children with ID generally show limitations in their level of cognitive development, and a slower learning curve for the skills they do develop (Katz & Lazcano-Ponce, 2008). This characteristic is also observed with regard to literacy skills (Erickson, Hanser, Hatch, & Sanders, 2009; Kaiser, Hester, & McDuffie, 2001). The level of reading comprehension in persons with ID varies widely. In a study including 19 literate adults (IQ 50-79), reading comprehension was found equal to that of typically developing children between 72 months and 114 months of age (Jones et al., 2006). Intervention studies evidenced that reading comprehension can be significantly improved in persons with mild to moderate ID by using direct and comprehensive instruction programs (Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2010; Ip & Lian, 2005; Van den Bos et al., 2007). However, the term 'reading comprehension' has been used in a broad sense and no clear distinction has yet been made between different aspects of reading comprehension, such as lower level (explicit) and higher level (implicit) reading comprehension. Studies attempting to identify specific cognitive or linguistic precursors to reading comprehension in children with ID are scarce. As a consequence, it is still unclear to what extent the construction of reading comprehension in children with ID differs from typically developing children.

Studies on early literacy achievement have shown that the attainment of decoding skill in children with ID is hindered by difficulties with phonological information processing (Iacono & Cupples, 2004; Ricketts, 2011). Nevertheless, with great effort, some children with ID do acquire decoding skills or achieve literacy by recognizing visual features of words (Allor et al., 2014; Conners, Atwell, Rosenquist, & Sligh, 2001). A large-scale American study indicated that around 32% of students with ID in grade 11 were able to decode at Grade 1 level, for 20% this was the highest level attained (Lemons et al., 2013). Teacher-ratings in Germany indicated that 32% of students with ID (age 6-21) were able to identify words without decoding them letter by letter (Ratz & Lenhard, 2013).

A few studies emphasized the importance of language comprehension for reading comprehension in children with Down syndrome. Although the language development of individuals with Down syndrome is not totally equal to the broader population of persons with ID (Chapman, 1997), these studies provide a starting point for studying the ID population. Roch and Levorato (2009) tested the applicability of the simple view of reading in 11- to 18-year-old individuals with Down syndrome. Language comprehension was a strong predictor for reading comprehension in this group, whereas decoding was not. Compared to a control group of typically developing children with equal reading comprehension skills, decoding was more advanced in children with Down syndrome while their language comprehension was weaker than in the control group. The authors concluded that reading comprehension in individuals with Down syndrome was constrained by their level of language comprehension (Roch, Florit, & Levorato, 2011; Roch & Levorato, 2009). Nash and Heath (2011) confirmed the importance of language comprehension skill for reading comprehension in thirteen 11- to 19-year-old individuals with Down syndrome. Furthermore, higher level reading comprehension, which required inference making, was more difficult for this group than would be expected based on their overall level of reading comprehension. Vocabulary and verbal working memory were of particular importance for inference making.

Relating to the lexical quality hypothesis (Perfetti, 2007), the vocabulary of individuals with ID is generally larger compared with typically developing children of the same mental age, but smaller than in typically developing children of the same chronological age (Facon & Bollengier, 2009). Although vocabulary seems to correlate with chronological age, other levels of language comprehension do not (Facon, Facon-Bollengier, & Grubar, 2002). Vocabulary instruction has been the focus of many studies on reading instruction in individuals with ID. These intervention studies have mainly addressed vocabulary in isolation, or embedded vocabulary instruction in a larger intervention program (Allor et al., 2014; Browder et al., 2006; Roberts, Leko, & Wilkerson, 2013). As a result, a causal link between vocabulary and reading comprehension has not yet been clearly established for this group.

Regarding cognitive skills, children with ID often have specific problems with reasoning and problem solving, even when they are compared to a typically developing control group matched on nonverbal intelligence (Facon & Nuchadee, 2010; Goharpey, Crewther, & Crewther, 2013). Especially problematic for individuals with ID is the understanding of spoken sentences with a complex syntactic structure (Jones, et al., 2006; Nation, 1999) or sentences that require inference making (Ezell & Goldstein, 1991). Limitations in any of these skills are likely to hinder the construction of a meaningful message from the individual elements of a text. In particular, problems in reasoning will be detrimental for achieving higher level inferential reading comprehension.

The Present Study

It is clear that children with ID struggle with reading comprehension and language comprehension, as well as several cognitive skills. Also, the final level of attainment in literacy is highly variable within the population. However, in the research so far, no attempt has been made to examine cognitive and linguistic predictors in children with ID who have learned the essentials of word decoding. Previous studies indicate that explicit comprehension requires a lower level of text processing than inferential comprehension (Nash & Heath, 2011, Kintsch & Van Dijk, 1978). In the present study, we therefore compared the levels of lower level (explicit) and higher level (implicit) reading comprehension in children with ID and typically developing children, and explored the contribution of linguistic skills to reading comprehension performance in children with ID.

A group of children with ID in the primary grades who had elementary decoding skill was compared to a control group of typically developing children from first grade whose word decoding skill was within the same range. Comparing groups of similar decoding level allowed for detection of differences in comprehension, with minimal interference of differences in decoding skill. Because cognitive skills have proven to be an influential factor in reading comprehension, nonverbal reasoning was first controlled for.

Two research questions were addressed: (1) How do children with ID compare to typically developing children who have the same elementary level of word decoding skill, with regard to lower level and higher level reading comprehension, and related cognitive and linguistic skills? (2) To what extent do linguistic skills contribute to lower level as well as higher level reading comprehension in children with ID who have attained an elementary level of decoding skill, when controlling for cognitive skill? Given the supposedly low levels of reasoning in the ID group, we expected lower scores on measures involving reasoning and inference making. Consequently, their higher level reading comprehension was expected to be more problematic than their lower level reading comprehension. Moreover, based on existing literature we expected that decoding, vocabulary and language comprehension would predict lower level as well as higher level reading comprehension, after controlling for nonverbal reasoning.

Method

Participants

Two groups of children were compared in the study: 49 children with ID who attended schools for special education in the Netherlands, and 21 typically developing children who attended regular education. Children were considered for participation when they had no visual or auditory impairments. The language spoken at home had to be Dutch only. Finally, for both groups, children were admitted to the study if they had a word decoding efficiency between 16 and 69 consonant-vowel-consonant (CVC) words per minute, which resembles the most common word speed during the second half of first grade in regular education in The Netherlands (Krom, Jongen, Verhelst, Kamphuis, & Kleintjes, 2010). All parents signed a consent form.

For the ID group, 65 schools were contacted of which 18 were willing to participate. The schools selected participants that fitted the criteria and contacted parents to request parental consent. The researchers only received information about students who had permission to participate. The final ID group consisted of 49 participants (28 male, 21 female; age between 111 and 155 months, M = 124.10 SD = 9.67), with mixed-etiology ID (IQ between 51 and 85, M = 62.71, SD = 8.62). Of students in the same age range, the participating schools labeled 41-47% as less proficient decoders than the selection criterion, 14 – 24% were labeled as better decoders.

The control group consisted of 21 children with a typical development who were in their second half of first grade (14 male, 7 female; age between 76 and 89 months, M = 82.33 SD = 3.93). They came from three schools for regular education. They were randomly selected from the population of 59 students that fitted the criteria for participation in these schools.

Measures

Children completed a number of tasks designed to tap cognitive and linguistic skills related to reading comprehension. For all tasks, the number of correct responses was used for further analysis.

Predictor Skills.

Word decoding. To re-assess decoding skill, the first reading card of the Drie-Minuten-Toets [Three-Minute-Test] was administered (Verhoeven, 1995). This card contains 150 monosyllabic words. The child was asked to read aloud the list of words quickly and accurately. The reported test score is the number of words read correctly in one minute. The reliability of the task is good, Cronbach’s alpha ranges from .86 (good) to .94 (excellent; Krom et al., 2010, Verhoeven & Van Leeuwe, 2008).

Vocabulary. A computerized version of the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Schlichting, 2005; Dunn & Dunn, 1997) was used to measure receptive vocabulary. In each trial, the child was required to choose one out of four pictures that best depicted a vocally presented word. The test was started at an appropriate difficulty for mental age, which was in this study the same for all participants. An adaptive testing paradigm then established receptive vocabulary of the child according to the number of correct responses given. The test was terminated when nine or more errors were made in a set of 12 items. Reliability (lambda-2-coëfficient) is excellent; reported between .93 and .97 for children between 7 and 13 years of age (Schlichting, 2005).

Language comprehension. The subtest Zinsbegrip 2 [Sentence Comprehension 2] from the Taaltoets Alle Kinderen [Language Test for All Children] (Verhoeven & Vermeer, 2001) measured understanding of explicit and implicit meaning relations within sentences. Explicit relation (i.e. anaphoric) comprehension required the child to understand semantic relationships within or between phrases. Understanding of implicit relationships (i.e. inferences) involved making presuppositions and the comprehension of modal verbs, which required the child to deduce the information implied by the presented sentence. The child chose one of three pictures that best matched the verbally presented sentence. In total, 42 items were presented. Cronbach’s alpha in first grade is good (.81; Verhoeven, & Vermeer, 2006).

Nonverbal reasoning. Non-verbal reasoning by analogy was measured by the Raven Coloured Progressive Matrices (RCPM; Raven, 1958). Children pointed to one out of six figures, selecting the figure that correctly completed an incomplete visual pattern. The test has 36 items in total. Split-half reliability for Dutch children aged between 6 and 9 has been reported between .82 and .87, which is adequate (Van Bon, 1986).

Reading comprehension.

Lower level reading comprehension. The ability to connect sentences by using explicit text cues was measured in the anaphora task. The child read four short stories of eight sentences that were selected from a larger reading comprehension task for first grade (Verhoeven, 1992). There were three multiple-choice questions about each text (each question has four answering possibilities). These questions

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referred to explicit story content and focused on the anaphora used in the stories. Cronbach's alpha in the present study was acceptable (.75).

Higher level reading comprehension. Four stories were selected from a reading comprehension test for first grade children to assess higher level reading comprehension (Reading Comprehension Test for First Grade; Aarnoutse, 1997). The child read four short stories of approximately ten sentences and answered three multiple-choice questions afterwards. The questions concerned sentence meaning (*which two sentences have the same meaning?*), making inferences about an event in the story (*why does John cry?*), and the main topic of the story (*what is the story about?*). The task measured the ability to make inferences about implicit information in the text. Cronbach's alpha was .75 in the present study, which is acceptable.

Procedure

The children participated during school hours in a separate, quiet room. The test battery took three sessions of approximately 30 minutes to complete, distributed over several days. The tests were administered by the first author and six trained undergraduate students Educational Science, who followed standardized instructions during testing. Children were tested by the same person during all sessions. They received a sticker after completing each task.

Data analysis

To allow comparison between groups, raw scores (i.e. the number of correct responses) were used. No correction for age or IQ was made before analysis. This way the absolute performance on each skill could be compared, leaving aside all other factors. Normality was checked by using the Shapiro-Wilk Test. All variables in both groups were normally distributed except higher level reading comprehension in the control group, due to an outlier. This was caused by the weakest reader in the control group scoring very poorly on reading comprehension. Since the requirement of decoding level was met, however, all scores were retained.

Results

Group comparisons

To answer the first research question, concerning how children with ID differ from the control group on the cognitive and linguistic skills, an independent samples t-test was conducted for each of the tasks. The groups differed significantly in age ($t(68) = 25.70$, $p < .001$, $d = 4.35$).

Table 1 shows the means and standard deviations for both groups, as well as the results of the independent samples t-tests. Children with ID scored below controls on language comprehension ($t(68) = 2.61$, $p = .011$, $d = 0.70$), nonverbal reasoning ($t(68) = 3.43$, $p = .001$, $d = 0.80$) and higher level reading comprehension ($t(68) = 3.21$, $p = .002$, $d = 0.84$). A paired samples t-test revealed that children within the ID group performed significantly weaker on higher level reading comprehension than on lower level reading comprehension ($t(48) = 2.58$, $p = .013$, $d = 0.37$), while children within the control group performed equal on both reading tasks ($t(20) = 0.31$, $p = .759$, $d = 0.07$).

Table 1.

Means, Standard Deviations and Contrast of Task Performance by the Control Group (n = 21) and Children with an Intellectual Disability (ID, n = 49)

	M (SD)			
	Control	**ID**	***t***	***d***
Word decoding	41.05 (13.07)	39.02 (14.27)	0.56	0.15
Vocabulary	93.00 (9.35)	93.06 (10.81)	0.02	0.01
Language comprehension	35.33 (3.23)	32.51 (4.46)	2.61**	0.69
Nonverbal reasoning	26.52 (3.98)	22.53 (5.41)	3.43**	0.80
Lower level reading comprehension	8.38 (2.60)	7.12 (2.86)	1.73	0.46
Higher level reading comprehension	8.57 (2.80)	6.18 (2.88)	3.21**	0.84

Note: Scores are not corrected for age or IQ.

* $p \leq .05$. ** $p \leq .01$.

Predictors of Reading Comprehension in children with ID

The second research question concerned the contribution of specific cognitive and linguistic predictors to reading comprehension in children with ID. Correlation coefficients among scores were first calculated to allow comparison of the strength of relationships between variables. The results are displayed in Table 2.

It can be seen in Table 2 that lower level reading comprehension was moderately correlated with word decoding ($r = .28$, $p = .048$), strongly correlated with language comprehension ($r = .40$, $p = .005$), and moderately correlated with nonverbal reasoning ($r = .32$, $p = .026$). Higher level reading comprehension was moderately correlated with nonverbal reasoning ($r = .29$, $p = .042$), and strongly correlated with lower level reading comprehension ($r = .61$, $p < .001$). In addition, vocabulary was found to moderately correlate with language comprehension ($r = .39$, $p = .008$). No significant correlation was found between vocabulary and the two reading comprehension tasks.

To identify the main predictors for reading comprehension, two hierarchical multiple linear regressions were conducted. The outcome variables were lower level reading comprehension and higher level reading comprehension. For both reading comprehension tasks, nonverbal reasoning was first entered into the model to control for cognitive skill. Second, decoding, vocabulary and language comprehension were entered into the model. The results are displayed in Table 3.
For lower level reading comprehension as well as higher level reading comprehension, a significant proportion of variance was explained by nonverbal reasoning. For lower level reading comprehension, linguistic skills explained unique additional variance. Decoding and language comprehension were both significant predictors in the model. For higher level reading comprehension, adding the linguistic predictors did not result in a significant increase of explained variance.

Table 2.

Correlation Coefficients between Task Performances of Children with Intellectual Disabilities (n = 49)

	1	2	3	4	5	6
1. Word decoding	-					
2. Vocabulary	.01	-				
3. Language comprehension	-.21	.39**	-			
4. Nonverbal reasoning	-.02	.10	.20	-		
5. Lower level reading comprehension	.28*	.25	.40**	.32*	-	
6. Higher level reading comprehension	.21	.05	.20	.29*	.61***	-

* $p \leq .05$. ** $p \leq .01$. *** $p \leq .001$.

Table 3.

Summary of Multiple Regression Analysis for Cognitve and Linguistic Skills Predicting Lower Level and Higher Level Reading Comprehension in Children with ID (n = 49)

Variable	***ΔR²***	***B***	***SE B***	**β**
Lower level reading comprehension				
Step 1				
Nonverbal reasoning	.11*	.17	.08	.33*
Step 2				
Word decoding		.08	.03	.38**
Vocabulary		.01	.04	.05
Language comprehension		.27	.09	.42**
	.26**			
Total R^2_{adj}	.31**			
Higher level reading comprehension				
Step 1				
Nonverbal reasoning	09*	.15	.07	.29*
Step 2				
Word decoding		.07	.03	.34*
Vocabulary		-.04	.04	-.15
Language comprehension		.18	.10	.28
	.14			
Total R^2_{adj}	.15*			

* $p \leq .05$. ** $p \leq .01$.

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Discussion

In the present study, children with ID and typically developing children who had a similar level of word decoding skill were compared on reading comprehension and predictor measures. The first question was how children with ID would compare to the control group of typically developing children with similar decoding skill, with respect to lower level and higher level reading comprehension, and cognitive and linguistic variables related to reading comprehension. Second, in the ID group we examined the contribution of linguistic skills to lower level and higher level reading comprehension, after controlling for cognitive skill.

With regard to the first research question, it was found that children with and without ID who had similar word decoding skill performed equally well on lower level reading comprehension and vocabulary. Note, however, that the children with ID were, on average, over 4 years older than the control group. Therefore, the equal scores do still express developmental lags in the ID groups in these domains. Additionally, the ID group performed significantly below the control group on language comprehension, nonverbal reasoning and higher level reading comprehension, which was in line with our expectations because these tasks required higher level reasoning skill and inference making. The greater difficulties of the ID group with tasks that required inference making compared to explicit comprehension concur with earlier findings in children with mild ID (Ezell & Goldstein, 1991; Nash & Heath, 2011).

Regarding the second research question, word decoding and language comprehension were found to be related to lower level reading comprehension after controlling for nonverbal reasoning. This indicates that the simple view of reading applies to children with ID with regards to lower level reading comprehension. The significant contribution of decoding to lower level reading comprehension in the present study is not in line with earlier findings in children with Down syndrome (Nash & Heath, 2011; Roch & Levorato, 2009). This might be explained by the difference in population characteristics. Individuals with Down syndrome tend to be fast word readers because they use visual recognition instead of decoding (Roch & Jarrold, 2012). None of the ID children in the present study had Down syndrome and they had all learned to read through word decoding in school. They were also younger than the participants in the studies of Roch and Levorato (2009). The current group can therefore be classified as less advanced in decoding skill and viewed as emergent readers with regard to the convergent skills model. In emergent readers, decoding is more important for reading comprehension than in proficient readers (Vellutino et al., 2007).

Despite a strong relationship between lower level and higher level reading comprehension, the influence of linguistic skills seem to be only predictive for lower level reading comprehension after controlling for cognitive skill. It could be speculated that higher level comprehension is strongly related to higher-order cognitive abilities related to executive functioning (cf. Locascio, Mahone, Eason, & Cutting, 2010; Sesma et al., 2009) which were not included in the present study. Unexpectedly, vocabulary was not related to reading comprehension which contradicts expectancies based on the lexical quality hypothesis and findings in children with Down syndrome (Nash & Heath, 2011; Perfetti, 2007). One reason might be that the words in our reading comprehension tasks were common words in the vocabulary of young children. Therefore, a larger vocabulary would not necessarily add to the level of comprehension of the particular texts in the present study. In line with this argument, Ouellette and Beers (2010) observed that, although vocabulary was a main predictor for reading comprehension in Grade 6, in Grade 1 it had no influence. Since the children in the present study were selected for decoding skills at first grade level, it is possible that their reading comprehension did not rely on vocabulary yet, but this pattern might change when decoding skill improves.

Nonverbal reasoning was related to reading comprehension in both lower level and higher level reading comprehension in children with ID. These results concur with findings in typically developing children that reading comprehension requires cognitive reasoning skills (Oakhill & Cain, 2012). The present results indicate that reasoning skill is not only involved in implicit inferential reading comprehension processes but also in explicit word-to-text integration processes.

Limitations and suggestions

The present study can only be seen as a first attempt to identify predictors of reading comprehension in children with ID, by including the main variables that have previously been identified as predictors for the normal population. A longitudinal study is necessary to establish the predictive value of the identified contributors over time. Also the use of a larger study group is preferred because of the variation in predictor and criterion measures within the general population of children with ID. The control group in the present study was small as well, which warrants caution in the interpretation of the results.

Undoubtedly, other cognitive and linguistic factors influence the level of reading comprehension than the predictors in the present study. In future studies, phonemic awareness and orthographic processing may be further investigated in relation to reading comprehension, since these are necessary for developing proper word decoding skills (Channell, Loveall, & Conners, 2013). Similarly, working memory and executive functioning are essential in several stages of the reading process, including reasoning and inference making, but are generally impaired in children with ID (Caretti, Belacchi, & Cornoldi, 2010; Schuchardt, Gebhardt, & Mächler, 2010; Sesma et al., 2009). Furthermore, we assessed vocabulary size but not the depth and organization of the vocabulary. Since deficits in associative memory appear to correlate with cognitive delay (Edgin, Pennington, & Mervis, 2010), the relationship between vocabulary and reading should also be explored further.

Implications

The present study points out the importance of decoding and language comprehension for lower level reading comprehension in children with ID, when controlling for nonverbal reasoning. No evidence has been found of a direct connection between vocabulary and reading comprehension in the present group. The simple view of reading (Hoover & Gough, 1990) therefore appears to be the best starting point when identifying predictors for reading comprehension in the ID population, next to an examination of their cognitive abilities.

In practice, the lack of an association between vocabulary and reading comprehension in the present group indicates that interventions focused on teaching individual words meanings may be insufficient for achieving comprehension of larger texts. Also, children with ID had more trouble with higher level text comprehension than children in the control group. Apparently inference making requires additional cognitive strategies which children which ID do not apply naturally. It is recommendable to focus on teaching the use of reading comprehension strategies in combination with general language comprehension and inferential reasoning. Teacher modeling has been successful in teaching students with mild ID to make inferences by using metacognitive strategies like the activation of prior knowledge and the use of question words while reading (Ip & Lian, 2009; Morgan, Moni, & Jobling, 2004). Children with poor reading comprehension seem to benefit most from an early intervention, using a reciprocal teaching approach (Snowling & Hulme, 2012). However, the process of word decoding should also be emphasized in reading education of children with ID. Although the children in the present study had considerable reading experience they were able to decode at a basic level only. Thorough understanding of word decoding principles through phonological awareness training (Channell, et al., 2013; Soltani & Roslan, 2013) or phonics instruction (Joseph & Seery, 2004) also may further improve their word decoding skill.

Conclusion

Based on the present findings, we may conclude that the reading profile of children with ID differs from that of children with a typical development, even when decoding skill is at a similar level. Children with ID who have learned how to decode written words seem able to perform explicit reading comprehension tasks. However, they demonstrate difficulties in understanding a text in spoken or written form, in particular when they are required to understand implicit meaning relations. Lower level reading comprehension abilities in children with ID could be explained by both cognitive and linguistic abilities, while higher level reading comprehension appears to rely less on linguistic abilities and more on cognitive skill. Problems regarding reading comprehension in children with ID thus seem to reflect a general deficit in the understanding of language due to a reduced use of logical reasoning strategies. The present study pointed towards the involvement of other cognitive skills in higher level reading comprehension that have yet to be ascertained. On the basis of these findings, it can be recommended that reading education for children with ID should highlight the instruction of reading comprehension strategies focusing on both explicit and implicit text meaning in addition to continuous support of basic word decoding skill.

Ch. 2

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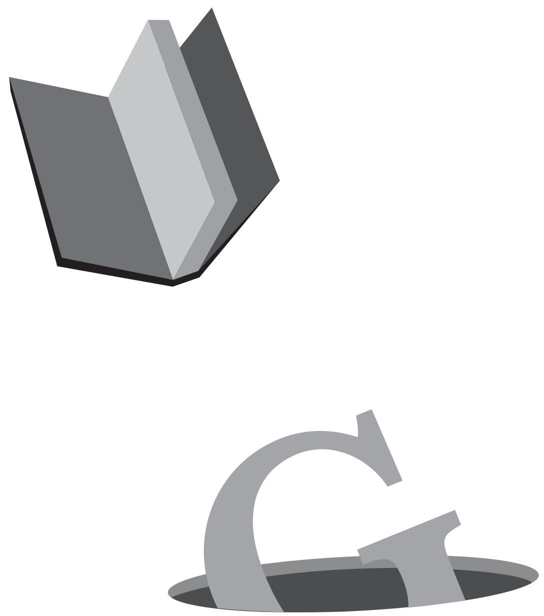

3.

Foundations of Reading Comprehension in Children with Intellectual Disabilities

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Abstract

Knowledge about predictors for reading comprehension in children with intellectual disabilities (ID) is still fragmented. The present study compared reading comprehension, word decoding, listening comprehension, and reading related linguistic and cognitive precursor measures in children with mild ID and typically developing controls. Moreover, it was explored how the precursors related to reading achievement. Children with mild ID and typical controls were assessed on reading, and linguistic (early literacy skills, vocabulary, grammar) and cognitive (rapid naming, phonological short-term memory, working memory, temporal processing, nonverbal reasoning) precursor measures. It was tested to what extent variations in reading comprehension could be explained from word decoding, listening comprehension and precursor measures. The ID group scored significantly below the control group on all measures. Word decoding speed in half the ID group was at or above first grade level. Reading comprehension in the ID group was related to word decoding, listening comprehension, early literacy skills, and temporal processing. The reading comprehension profile of children with mild ID strongly resembles that of early readers. The simple view of reading pertains to children with ID, with an additional impact of early literacy skills and temporal processing.

The simple view of reading states that reading comprehension is essentially a product of word decoding and listening comprehension (Hoover & Gough, 1990). For children with intellectual disabilities (ID), learning to read is less straightforward than for typically developing children, and reading comprehension causes additional difficulties. A large proportion does acquire literacy to some degree, but the reading level differs widely (Jones, Long, & Finlay, 2006; Lemons et al., 2013; Ratz & Lenhard, 2013). Although recent studies in children with ID did focus on predictors of word decoding and predictors of reading comprehension (Nash & Heath, 2011; Soltani & Roslan, 2013), a comprehensive model has not yet been established. In addition, it is unclear to what extent reading-related abilities such as working memory, rapid naming, and vocabulary contribute to literacy performance in this population. Recent studies have shown that the simple view of reading applies to individuals with Down syndrome (Nash & Heath, 2011; Roch, Florit, & Levorato, 2011), however, these results may not generalize to other etiologies. The present study aimed to investigate to what extent the reading comprehension abilities of children with mixed-etiology mild ID can be explained from their word decoding and language comprehension, and relevant linguistic and cognitive precursor measures.

Typical reading development

In typical readers, it is known that a combination of word decoding and listening comprehension determines the level or reading comprehension (Hoover & Gough, 1990).

For word decoding the reader must merge letter-sound combinations into words, which requires a basis of underlying linguistic skills. First, the reader must be aware of the different sounds and sound clusters within speech and be able to manipulate them (phonological awareness). Second, the reader must know the connection between speech sounds and their corresponding letters (letter knowledge). This combination of skills (to be called ‘early literacy skills’ from this point forward) are closely related to the attainment of word decoding in orthographies of different levels of transparency (Melby-Lervåg, Lyster, & Hulme, 2012). Phonological representations in memory are further refined when vocabulary expands (Walley, Metsala, & Garlock, 2003). Additionally, vocabulary knowledge supports word identification (Perfetti, 2007). Language-related cognitive skills are necessary for a fluent word decoding process. Rapid naming is related to word reading fluency, as it reflects processing speed and the level of integration of linguistic and perceptual reading-related processes (Norton & Wolf, 2012). In addition, phonological short-term memory capacity is necessary for blending letters into a word, and temporal processing to enable speech perception, which is instrumental in the development of phonological awareness (Georgiou, Torppa, Manolitsis, Lyytine, & Parrila, 2012; Huss, Verneij, Fokster, Mead, & Goswami, 2011; Malenfant et al., 2012; Perez, Majerus, & Poncelet, 2012).

Once children have reached a basic level of word decoding, they learn to incorporate written words into meaningful sentences and text. For beginning readers, word decoding is the main determinant of reading comprehension. Over time, word decoding becomes an automatic process and reading comprehension becomes mainly dependent on language comprehension (Vellutino, Tunmer, Jaccard, & Chen, 2007). Listening comprehension is the second element of the simple view of reading. It requires vocabulary for knowledge of word meanings (Lee, 2011) and grammar comprehension for sentence comprehension (Kintsch & Rawson, 2005). For comprehension of texts, working memory and reasoning skills are needed for text integration and to perform the more complex tasks such as inference drawing and use of reading strategies (Fuchs et al., 2012; Seigneuric & Ehrlich, 2005). Temporal processing is necessary for speech perception, grasping the order of phonemes and words, and detecting the prosodic patterns in spoken language (Gordon, Jacobs, Schuele, & McAuley, 2015; Malenfant et al., 2012). Finally, recent studies have pointed out that vocabulary is also an independent predictor for reading comprehension, next to decoding and language comprehension (Protopapas, Mouzaki, Sideridis, Kotsolakou, & Simos, 2013).

Reading in children with mild ID

Children with mild ID have severe delays in working memory (Van der Molen, Van Luit, Jongmans, & Van der Molen, 2007) and early literacy skills, which hinders the development of word decoding, and reading comprehension accordingly (Channell, Loveall, & Conners, 2013; Jones et al., 2006). In addition, Levy (2011) found in adolescents with moderate to borderline ID of different etiologies that the general level of cognitive capacity affects the reading level on top of language-related predictors.

Several large studies have inventoried the word decoding levels of children with ID through teacher reports. One study included 1629 German students of 6- to 21-year-old with ID, regardless of etiology. Regarding the 529 students who were classified as having mild ID, their teachers reported that 35.8% decoded by deliberate, letter-by-letter decoding, and 59% was able to read by direct orthographic word recognition (Ratz & Lenhard, 2013). In a second study, scores on a curriculum-based reading measures were used to determine word decoding levels among 3811 American

students of 8 to 21 years old who had mild to moderate ID. The benchmark for first grade word decoding level was obtained by 33.6 % on average, with the percentage increasing by age (Lemons et al., 2013).

Although children with mild ID lag behind in the development of reading, the underlying predictors seem, to a certain extent, similar to those in typically developing children. Early literacy skills (Dessemontet & De Chambrier, 2015) and rapid naming (Barker, Sevcik, Morris, & Romski, 2013; Soltani & Roslan, 2013) are prominent predictors for word decoding in mild ID. Phonological short-term memory is an indicator in some studies (Conners, Atwell, Rosenquist, & Sligh, 2001), but in other studies was not predictive when phonological awareness was controlled for (Soltani & Roslan, 2013). Compared to typically developing children, however, the word decoding level of children with mild ID appears to be more strongly related to cognitive skills such as nonverbal reasoning and temporal processing (Van Tilborg, Segers, Van Balkom, & Verhoeven, 2014).

Reading comprehension has been studied less frequently in individuals with ID than word decoding, and indications of reading comprehension levels are rarely reported. One study among 19 literate adults with mild ID, revealed a reading comprehension level equal to typically developing children of 72-114 months (Jones et al., 2006). Studies regarding the predictors of reading comprehension in individuals with ID are also sparse. One recent study on 129 children with mild or moderate ID of unspecified etiology found that early literacy skills (phonological awareness and letter knowledge) at age 6-8 were predictive for reading comprehension one and two years later, when controlling for IQ, age, expressive vocabulary, native language, and school placement (Dessemontet & De Chambrier, 2015). Word decoding and listening comprehension were not taken into account in this analysis. Several other studies have found results that provide more direct support the simple view of reading. In a group of 49 children with mixed-etiology mild ID who had basic word decoding skills, word decoding was the primary predictor for reading comprehension, after controlling for nonverbal reasoning. A significant relationship was also found with listening comprehension, but not with vocabulary (see Chapter 2). In a longitudinal study of ten adolescents with Down syndrome with more advanced word recognition skills, the development of reading comprehension was mainly determined by listening comprehension, while the relationship with word decoding was not significant (Roch et al., 2011). In a similar vein, in a cross-sectional study of 13 participants with Down syndrome the correlation between word decoding and reading comprehension was only marginally significant, while strong connections were found between reading comprehension and vocabulary, which was considered indicative for language ability (Nash & Heath, 2011). Indeed, it has been shown that the comprehension process in text reading is problematic in persons with ID, in particular when the meaning of the text is ambiguous and requires inference making (Nash & Heath, 2011; Tavares, Fajardo, Ávila, & Salmerón, 2015). Vocabulary does not seem to be a direct predictor for reading comprehension in children with mild ID (Dessemontet & De Chambrier, 2015), but only to relate through language comprehension (Verhoeven & Vermeer, 2006a).

Information on additional predictors for reading comprehension in individuals with mild ID is limited, and is mainly based on intervention studies. These studies have yielded positive effects of comprehensive reading instruction that incorporated strategy instruction on literacy outcomes, including reading comprehension, (e.g. Allor, Mathes, Roberts, & Cheatham, 2014). A recent longitudinal study regarding 69 adolescents with velo-cardio-facial syndrome with borderline ID indicated that attention, self-monitoring, and working memory predicted reading comprehension next to vocabulary and word decoding (Antschel, Hier, Fremont, Faraone, & Kates, 2014). Other studies on individuals with mild or moderate ID have identified specific problems in reasoning and working memory that may be related to reading comprehension problems (Carretti, Belacchi, & Cornoldi, 2010; Numminen, Service, & Ruoppila, 2002). It is not yet certain to what extent a direct connection between cognitive factors and reading comprehension exists for children with mild ID.

The present study

Although predictors for decoding and, to a lesser degree, reading comprehension have been identified for children with mild ID, this knowledge is still fragmented. Studies have focused primarily on linguistic precursors of reading, and several studies concerned specific populations. The present study aimed to incorporate reading comprehension, word decoding, language comprehension, and relevant linguistic and cognitive precursor measures in one model for a non-specific group of children with mild ID. Studies so far seem to suggest that literacy development in children with mild ID follows a similar path as in typically developing children, but cognitive weaknesses may cause additional difficulties in processing linguistic information and semantic relations. In the present study, early literacy skills, vocabulary and grammar comprehension were taken as linguistic precursors (Dessemontet & De Chambrier, 2015; Nash & Heath, 2011), and rapid naming, phonological short-term memory, working memory, temporal processing and nonverbal reasoning as cognitive precursors (Barker et al., 2013; Gordon et al., 2015; Nash & Heath, 2011).

First, linguistic and cognitive skills in children with mild ID were compared to scores of typically developing children who had reading instruction for the same number of years, to investigate the extent of developmental delay in these areas. Second, the variation in word decoding level within the ID group was inventoried to explore to what extent they had achieved this first step in the reading process. Finally, we aimed to identify the main predictors for reading comprehension in children with mild ID, and in children with typical development. Although much is known about reading development in typically developing children, information on predictors in children with mild ID has been fragmented and an overarching framework has not yet been established for this group. Combining the available information so far will provide insight in the coherence of these elements. The participants in the present study were monolingual Dutch speakers, which has a transparent orthography. This enabled examining the role of word decoding, rather than word recognition, in early literacy (Seymour, Aro, & Erskine, 2003; Share, 2008).

We expected significantly lower scores in children with mild ID compared to a typically developing control group on all measures (Channell et al., 2013; Lemons et al., 2013; Numminnen et al., 2002). Second, in line with the data from Lemons et al. (2013), we expected that most children with mild ID would show word decoding skills at the lower grade levels. Furthermore, we expected to find that the simple view of reading also pertains to children with mild ID of mixed etiologies, similar to Roch et al. (2011). However, because of the additional cognitive deficits that are associated with ID, we also anticipated that direct effects of one or more precursor measures such as early literacy skills and working memory might occur (Dessemontet & De Chambrier, 2015; Nash & Heath, 2011).

Method

Participants

The present study was conducted in the Netherlands, and included a group of children with ID and a control group of children with typical language development. Children with ID were all in schools for special education, as is true for the majority (90%) of children with ID in The Netherlands (Henkens, 2008). No children with special needs were included in the control group. For both the ID group and the control group, inclusion criteria were normal hearing, eyesight that was normal or corrected to normal, and a monolingual Dutch-speaking home environment. There were no requirements with regard to the reading level of the children. All participants in the study had had three years of reading instruction. However, in the

Netherlands, regular literacy instruction for children with ID generally starts one year later than for children in regular education (Van der Laan, 2006). In order to compare reading achievement in the ID group and the control group, the ID group in the present study was one year older than the control group so that the groups were of similar *instructional* age.

Since the Dutch language has a transparent orthography, reading instruction typically starts with decoding orthographically transparent words. This is followed by teaching word recognition of more opaque words. While the reading of short narratives begins soon after the start of word decoding instruction, purposeful instruction in reading comprehension (focusing on reading strategies) is generally introduced in second grade of Dutch education, when word decoding is sufficiently developed (Seymour et al., 2003). The approach to literacy learning for children with ID is not clearly defined. In most cases, reading education for children with ID is similar regular methods, supported with visual aids such as photographs, visual symbols, and iconic hand gestures representing words and letters. Much attention goes out to word decoding, while reading comprehension generally receives less attention. There is little instruction in reading strategies, and reading comprehension is mostly stimulated by independent bookreading (Van der Laan, 2006).

For the control group, 5 schools for regular education responded to a general invitation that was sent out to 49 randomly selected schools spread across The Netherlands. These schools did not include children with ID. All children in third grade who met the general inclusion criteria were asked to participate. The final control group consisted of 84 third-grade students (35 girls, 49 boys). Age ranged between 99-126 months (M = 108.02, SD = 5.77). The weighted average social-economic status in the catchment area of the participating schools was in line with the average level in The Netherlands (Sociaal en Cultureel Planbureau [SCP], 2012).

For the ID group, 20 schools responded to an invitation that was distributed among 66 schools for special education in The Netherlands. The participating schools were asked to select participants that fitted the criteria (age between 108 and 138 months, IQ 50 – 80) and contacted parents to request parental consent. The researchers only received information about students who had permission to participate. The final ID group consisted of 81 children (29 girls, 52 boys) with ID of mixed etiologies (IQ 50 – 80, M = 60.38, SD = 7.20). The age of the children ranged between 107-137 months (M = 121.27, SD = 5.83). The participants in the ID group did generally not live close to their schools and therefore we could not estimate socio-economic status based on the location of the school. However, by means of a questionnaire the education level of the father could be determined for 49 participants. The distribution of these education levels was analogous to national average numbers in The Netherlands (Centraal Bureau voor de Satistiek [CBS], 2013).

Measures

Participants completed several tasks that measured cognitive and linguistic skills related to reading comprehension. For reading comprehension and temporal processing we used tests that were unpublished, but validated in earlier studies. The reading comprehension test was based on existing standardized tests for reading comprehension. The temporal processing task has been repeatedly used for diagnostic studies in Dutch children who were deaf. All other tasks were standardized tasks originating from established Dutch diagnostic tests. Unless specified otherwise, the reported test score is the number of correct responses on a task.

Reading comprehension in the ID group was measured using two reading tasks, appropriate for the first grade of regular education. Earlier studies have shown this to be the highest achievable reading level for the majority of children with ID within the current age range (Van Wingerden et al., 2014; see also Lemons et al., 2013). These tasks were taken individually, as part of the general test sessions. In each task, children read four stories of 8-10 sentences that they read silently to themselves. The stories were followed by three multiple-choice questions, which they also read independently. The first task focused on anaphora (explicit anaphoric story content; part of Verhoeven 1992). The second task focused on inferences (implicit story information; part of Aarnoutse 1997). Reliability of both tasks was good (Cronbach's alpha = .85 for the anaphora task and .75 for the inferences task). Reading comprehension in the control group was tested in the classroom at grade-appropriate level (Grade 3) using a standardized reading comprehension task that is part of the national Dutch student monitoring system (Cito, 2007). Two series of 25 multiple-choice items were administered by the teacher. The second series was either slightly easier, or slightly more difficult than the first series, depending on the score that was obtained on the first series. The raw scores were then converted to a norm score. For Grade 3, the national average norm score is 25.5, SD = 12.5 (Keuning, Hilte, & Weekers, 2014). The reading comprehension level of all children in the control group was determined at least at Grade 2 level according to their scores on this reading comprehension task. Cronbach's alpha = .85, which is good (Feenstra, Kleintjes, Kamphuis, & Krom, 2010).

Decoding was tested with two timed single-word reading tasks; one part measured word decoding and the second part measured pseudoword decoding in the same manner. In both cases, the outcome measure was the number of words read correctly within the available time. *Word decoding* was measured using two word lists of different orthographic complexity. For each card, participants read as many words as possible in 1 minute. The test is part of the national Dutch student monitoring system (Verhoeven, 1995). The reported test score is the total number of words read correctly for the two cards combined. Cronbach's alpha ranges from .86 (good) to .94 (excellent; Krom, Jongen, Verhelst, Kamphuis, & Kleintjes, 2010). *Pseudoword decoding* was a subtest of the Dutch Screening test for Specific Language Impairment (Verhoeven, 2005). Two pseudoword lists of different orthographic complexity were presented to the participants. Here the number of correctly read words within 2 minutes was calculated for each card and these scores were added. Cronbach's alpha reliability is above .85.

Listening comprehension was tested by reading aloud five short stories from the Text comprehension subtest of the Clinical Evaluation of Language Fundamentals 4-NL (Kort, Jongen, Verhelst, Kamphuis, & Kleintjes, 2008). All stories were between 67 and 115 words in length and were appropriate for children between 7-10 years. After each story, five open-ended questions were asked that either directly referred to the story content, or required additional reasoning. Cronbach's alpha is good (between .71 and .74 for this age category; Egberink, Vermeulen, & Frima, 2014).

Early literacy skills in the ID group were measured using a computer version of the Diagnostic Instrument for Emerging Literacy (Instituut voor Orthopedagogiek, 2004). Four competences were assessed: rhyme, synthesis, deletion, and letter knowledge. Three of the tests had 15 multiple-choice items where one out of three pictures had to be selected. They included high-frequency monosyllabic Dutch words. In the *rhyme*-task, participants selected the picture representing a word rhyming with an aurally presented stimulus. In the *synthesis* task, participants heard a string of letters that spelled the target word. In the *deletion* task, participants heard a stimulus word of which they then had to delete a certain phoneme, which resulted in a new word. In the *letter knowledge* task, four letters were shown on the computer screen and the participants had to select the letter or digraph corresponding to an auditory stimulus. This task contained 34 items. Reliability of each task is good (Cronbach's alpha = .78; Egberink et al., 2014). The control group only completed the deletion task, as they were expected to score at ceiling level on the rhyme, synthesis and letter knowledge tasks (cf. Vloedgraven & Verhoeven, 2007).

Vocabulary was tested using the Dutch version of the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Schlichting, 2005). During each trial, the participant chose one out of four pictures that best corresponded to a verbally presented word. A start set was chosen that was appropriate for children of 5;6 – 6;5 years of age. Item difficulty increased as the participant progressed through the stimulus sets, until the ceiling level of the participant was reached. Reliability (lambda-2-coëfficient) is excellent; reported between .93 and .97 for children between 7 and 13 years of age.

Grammar comprehension was tested using a multiple choice computer task comprising aurally presented sentences (Verhoeven & Vermeer, 2001). The 42 sentences required understanding of explicit and implicit meaning relations within each sentence. Participants selected one of three pictures that corresponded most to the meaning of the sentence. Cronbach's alpha in first grade is good (.81; Verhoeven & Vermeer, 2006b).

Rapid naming included five different line drawings (representing a duck, glasses, shoe, house and comb), repeated in random order along four columns of 30 items (120 pictures in total), presented on one page (Verhoeven, 2005). The participant was asked to name the pictures in the presented order as quickly and accurately as possible, during one minute. Reliability is excellent (Cronbach's alpha = 0.95).

Pseudoword repetition comprised a series of 40 aurally presented pseudowords, which the participant was asked to repeat (Verhoeven, 2005). Phonemic complexity of the words increased, after five consecutive errors the task was terminated. Reliability is excellent (Cronbach's alpha = .94).

Working memory was tested using the Digit Span subtest of the Wechsler Intelligence Scale for Children – Dutch version (WISC-III-NL; Kort et al., 2005). Participants repeated strings of digits that increased in length, until a ceiling level was reached. First the digits were repeated in the same order, later the order had to be reversed. Reliability is acceptable (Cronbach's alpha = .64).

Temporal processing was measured using the Rhythm Test (Van Uden, 1983). Participants were asked to repeat rhythms of increasing complexity by tapping on the table with a pencil. After two repetitions immediately after demonstration by the experimenter, participants repeated each rhythm five more times. The test continued until all a ceiling level was reached or until all 15 items had been presented. No reliability scores are known for this task.

Nonverbal reasoning was tested using Raven's Coloured Progressive Matrices (RCPM; Raven, 1958). Children chose one out of six puzzle pieces that best completed a larger visual pattern. All 36 items were administered. Split-half reliability for Dutch children aged between 6 and 9 has been reported between .82 and .87, which is adequate (Van Bon, 1986)

Procedure

The research was approved by a national institute for individuals who are deaf, or have other disabilities that hinder communication. Active parental consent was obtained by the schools for all participating students, before the testing started. Children were tested during school hours in a separate room. For the ID group, testing took 120 minutes, divided over four sessions of 30 minutes. Tests were administered by the first author and two undergraduate students of Educational Science who had received extensive training to follow standardized instructions. For the control group, testing required a total of 90 minutes, divided over two sessions of 45 minutes. This group was tested by the first author and four undergraduate students. Reading comprehension in the control group was tested beforehand by the teacher in a classroom setting as part of the national student monitoring program.

Data Analysis

Missing data occurred on 0.54% of the total number of data points. These data points were filled using multiple imputation. Pooled results are reported. To answer the first research question, t-tests for independent samples were performed on all individual tasks that overlapped between the two groups. Second, to explore word decoding in the ID group, scores were divided into categories of reading level, according to national norms for students in first grade of primary school. Finally, the strength of relations between constructs was explored for the both groups. We calculated z-scores for all scores to reduce any non-normality in the data. For the ID group, based on a confirmatory factor analysis composite scores were created, by adding the z-scores of these measures: reading comprehension consisted of anaphoric and inferential reading comprehension (R^2 = .88), early literacy skills consisted of rhyme, synthesis, deletion and letter knowledge (R^2 = .54), and decoding consisted of word decoding and pseudoword decoding (R^2 = .98). The composite scores were then treated as measured variables in further analysis. Correlation coefficients were calculated to examine the strength of relations between all measures. Structural Equation Modeling (SEM) analyses in LISREL were used to evaluate the relationships between cognitive and linguistic factors in relation to reading comprehension in the ID group. The model fit was evaluated according to six indices: the χ^2:df ratio should be lower than 2:1, χ^2 significance should exceed .05, the Non Normative Fit Index (NNFI), Comparative Fit Index (CFI), Adjusted Goodness of Fit Index (AGFI) and Goodness of Fit Index (GFI) should exceed .90. The Root Mean Square Error of Approximation (RMSEA) should be below .05 (Baumgartner & Homburg, 1996; Ping, 2004).

Results

The means and standard deviations for all scores are displayed in Table 1. For the control group, several scores are left blank in this table, as these tasks were not administered in this group. First, to compare the mean scores in the ID group and the control group, t-tests for independent samples were performed on all individual tasks that overlapped between the two groups (Table 1). The ID group scored below the control group on all tests; all group comparisons were significant ($p < .001$) and effect sizes were high on all measures (between d = 1.32 for listening comprehension and d = 5.32 for word decoding).

In exploration of the decoding level of the ID group, the large standard deviation of decoding skill in this group is interesting (Table 1). To gain additional insight into the distribution of decoding abilities in this group, we used the standards provided with the word decoding task (Jongen & Krom, 2009). Of the 81 participants in the ID group, 43 participants scored in the lowest 25% range of typically developing first-graders (less than 29 words read over both reading cards, M = 9.23, SD = 8.80), 13 participants fell in the middle 50% category (between 30 – 61 words read, M = 41.92, SD = 8.89) and 25 participants were classified as the top 25% (more than 62 words read, M = 110.88, SD = 37.79) according to norms for first grade. A small subgroup of 6 participants with ID had a word decoding level that was comparable to the average word decoding level in third grade. In the control group the word decoding level was at or above third grade level for all children.

Finally, relationships between variables were explored further for the ID group and the Control group. Note, however, that a different measure for reading comprehension was used in the two groups, due to large differences in reading level. For the same reason, not all measures for early literacy skills were included in the analyses for the control group. In Table 2, correlations are shown between cognitive predictors and the composite scores that were constructed for reading comprehension, early literacy skills and decoding. As can be seen in Table 2, for the ID group most predictor and outcome variables were significantly correlated except temporal processing and listening comprehension. For the

Table 1.

Means, standard deviations and group comparisons for children with an intellectual disability (ID; n = 81) and the control group (n = 84)

	Max. score	Exp. score	ID *M*	ID *SD*	Control *M*	Control *SD*	*t*	*d*
Reading comprehension								
Grade 1 Anaphora	12	12	5.27	3.72	[b]			
Grade 1 Inferences	12	12	5.00	3.07	[b]			
Grade 3	-	25.5	[a]		28.00	14.86		
Decoding								
Word decoding	-	392	45.85	50.26	168.30	32.08	18.58**	5.32
Pseudoword decoding	-	372	55.63	71.56	212.13	55.14	15.71**	2.47
Listening comprehension	20	18	11.17	6.24	17.73	3.43	8.22**	1.32
Early literacy skills								
Rhyme	15	15	10.99	3.22	[b]			
Synthesis	15	15	12.52	2.92	[b]			
Deletion	15	15	6.75	3.16	13.90	1.90	17.36**	2.77
Letter knowledge	34	34	30.86	5.80	[b]			
Grammar	42	40	29.85	6.78	38.55	2.44	10.78**	1.73
Vocabulary	-	125	88.16	16.09	111.61	8.76	11.56**	1.83
Rapid naming	120		45.12	14.41	61.26	9.58	8.44**	1.33
Pseudoword repetition	40	30	12.40	9.32	28.05	7.10	12.10**	1.91
Working memory	30	13	7.06	2.38	12.23	2.39	13.92**	2.18
Temporal processing			27.57	36.73	114.85	53.97	12.18**	1.89
Nonverbal reasoning	36	31	19.94	6.11	31.20	3.43	14.54**	2.30

Note. Max. score = Maximum score on the test, if applicable. Exp. Score = expected score on test, given mean age of the participants.

[a] Assumed to be at floor level.

[b] Assumed to be at ceiling level.

** $p < .01$

control group, reading comprehension was significantly correlated were with all linguistic predictors, phonological short-term memory, and nonverbal reasoning. Decoding was correlated with early literacy skills, rapid naming, and working memory. Listening comprehension was correlated with grammar and vocabulary.

Table 2.

Correlations between predictor and outcome variables within the ID group (*n* = 81) below the diagonal and the Control group (*n* = 84) above the diagonal

	1[a]	2[b]	3	4	5	6	7	8	9	10	11
1. Reading comprehension	-	.26*	.38**	.48**	.14**	.41**	.06	.36**	.21	.13	.33*
2. Early literacy skills	.66**	-	.32**	.18	.20	.10	.22	.20	.24*	.30**	.20
3. Decoding	.75**	.54**	-	.13	.05	-.07	.31**	.11	.25*	.13	-.13
4. Listening comprehension	.49**	.50**	.33**	-	.30**	.31**	.04	.17	.07	.07	.18
5. Grammar	.58**	.72**	.43**	.66**	-	.23	.28*	.19	.17	.14	.43**
6. Vocabulary	.47**	.61**	.40**	.71**	.75**	-	-.08	.03	.03	-.13	.33**
7. Rapid naming	.54**	.53**	.55**	.33**	.47**	.41**	-	-.17	.21	-.04	.20
8. Pseudoword repetition	.46**	.54**	.41**	.45**	.43**	.51**	.22*	-	.12	.37**	.14
9. Working memory	.55**	.70**	.50**	.48**	.61**	.56**	.51**	.49**	-	.37**	.25*
10. Temporal processing	.46**	.45**	.30**	.20	.31**	.31**	.23*	.48**	.46**	-	.12
11. Nonverbal reasoning	.50**	.57**	.30**	.42**	.61**	.49**	.42**	.33**	.54**	.43**	-

Note. * $p \leq .05$. ** $p \leq .01$.

[a] different reading comprehension tasks for each group.

[b] For the control group, only the deletion task was used.

With respect to the path model, structural equation modeling was used to identify the main predictors for reading comprehension in the ID group and separately for the Control group. We emphasize that the resulting models cannot be compared directly, due to a difference in the outcome measure. Separate path analyses were performed for the two groups. Both analyses started with the known predictors for reading comprehension (decoding and listening comprehension), decoding (rapid naming and early literacy skills) and listening comprehension (vocabulary and grammar). To identify additional predictors, the remaining cognitive and linguistic precursor measures were then added to the model of known predictors for each linguistic skill (decoding, listening comprehension, and reading comprehension) separately.

For the control group, listening comprehension was related to the initial predictors only. None of the precursor measures reached significance in predicting decoding. For reading comprehension, vocabulary and nonverbal reasoning were additional predictors next to decoding and listening comprehension. The final model (Figure 1) showed that reading comprehension was predicted by decoding, listening comprehension, vocabulary and nonverbal reasoning (R^2 = .38) and listening comprehension was predicted by vocabulary and grammar (R^2 = .15). The fit of the final model was acceptable ($\chi^2(3)$ = .5.43, p = .143, NNFI = 0.83, CFI = .97, AGFI = .86, GFI = .98, RMSEA = .099).

For the ID group, no additional cognitive precursor measures reached significance on top of the initial predictors for word decoding and listening comprehension. Regarding reading comprehension, vocabulary was not significantly related to reading comprehension and was excluded from the model. Conversely, temporal processing and early literacy skills were significant additional predictors in the model, next to decoding and listening comprehension. Next, the findings above were combined into one overarching model. The final model (Figure 2) showed that reading comprehension was predicted by temporal processing, early literacy skills, decoding and listening comprehension (R^2 = .69). Decoding, in turn, was predicted by early literacy skills and rapid naming (R^2 = .38), and listening comprehension was predicted by vocabulary and grammar (R^2 = .54). The fit of the final model was excellent ($\chi^2(10)$ = 5.11, p = .884, NNFI = 1.02, CFI = 1.00, AGFI = .94, GFI = .98, RMSEA = .00).

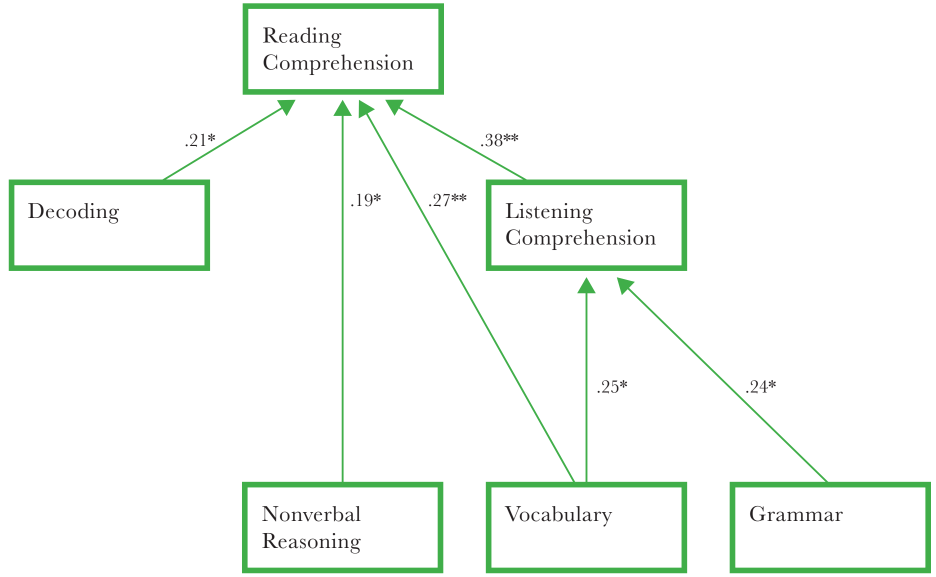

Figure 1.
Path model for cognitive and linguistic predictors of reading comprehension in the control group.

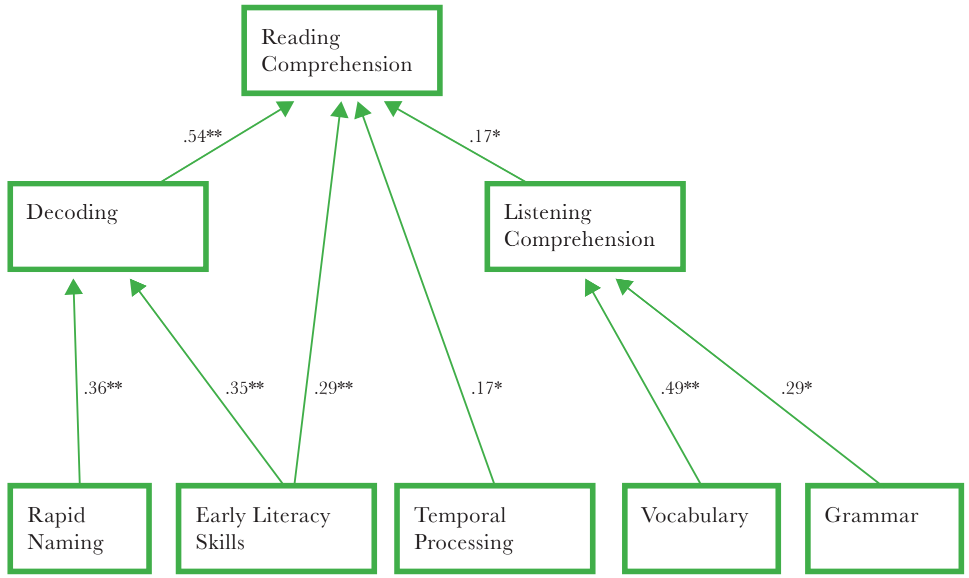

Figure 2.
Path model for cognitive and linguistic predictors of reading comprehension in the ID group.

Discussion

The aim of the present study was to identify a model of cognitive and linguistic predictors for literacy acquisition in children with mild ID. The results showed that children with ID lag severely behind on cognitive as well as linguistic skills compared to typically developing children of a similar instructional age. Approximately half of the ID group had a single-word decoding speed at or above Grade 1 level. Finally, the structural models for this group indicate that the simple view of reading applied in both groups. In the control group, an additional contribution of vocabulary and nonverbal reasoning was found, while for the ID group additional connections were found with early literacy skills and temporal processing. Cognitive as well as linguistic predictors connected to decoding and reading comprehension in children with mild ID.

Regarding the first research aim, the difference in test scores between children with ID and typically developing children was expected, given the existing literature. The present results are in line with earlier findings that individuals with ID lag behind in cognitive and linguistic development (Jones et al., 2006; Lemons et al., 2013; Numminen et al.; 2002). Besides a considerable arrear in reading comprehension, the ID group scored significantly lower on all predictor variables compared to a group of typically developing children who had the same instructional age, which indicates a general delay in literacy-related skills. Although the groups could not be compared in reading comprehension and several early literacy skills, it was clear that the ID group did not reach ceiling level on standardized tests that were designed for children who are over three years younger. Early literacy was not fully developed within the ID group, despite three years of reading instruction. Their average level of word decoding skill reflects this delay. Mean reading comprehension scores indicate that the level of reading comprehension is below Grade 1 level for most participants in the ID group. In fact, very few participants obtained a maximal score on this test, even though a large proportion had accomplished a word decoding efficiency at or above the average decoding levels for first-graders. These results illustrate a mismatch between decoding skill and reading comprehension, similar to children with Down syndrome and 'poor comprehenders', who struggle with comprehending both spoken and written language while decoding skills are fluent (Nation, Cocksey, Taylor, & Bishop, 2010). Reading education for children with ID could benefit from existing knowledge about these groups, whose reading comprehension skills have been studied more extensively.

Secondly, the decoding level of the children with ID was further explored. In the current study, 47% of the participants with mild ID attained a single-word decoding speed at or above the pace of an average first-grade student. This percentage is slightly lower the findings of Ratz and Lenhard (2013) in German, who used teacher questionnaires to determine that 59% of children with mild ID had reached an orthographic reading level. However, the difference in assessment methods and in age range of the participants hinders a direct comparison between the two studies. The percentage is higher than was found in American students using standardized tests (Lemons et al., 2013), which may be partly due to a difference in orthography (Seymour et al., 2003) and partly to differences in the age range. Still, the present results concur with earlier findings that reading skills among children with ID vary widely, even within a relatively small range of age and IQ. The predictors in the present study accounted for a significant amount of this variation, but not all could be explained. It is important to keep this variability in mind when looking at general developmental patterns in persons with ID.

Finally, reading comprehension and its predictors in children with ID and the control group were modeled. Early literacy skills, vocabulary and grammar comprehension were taken as linguistic precursors (Dessemontet & De Chambrier, 2015; Nash & Heath, 2011), and rapid naming, phonological short-term memory, working memory, temporal processing and nonverbal reasoning as cognitive precursors (Barker et al., 2013; Gordon et al., 2015; Nash & Heath, 2011).

In the control group, reading comprehension was predicted by decoding, language comprehension, and vocabulary in accordance with the simple view of reading (Hoover & Gough, 1990), and several recent additions (Protopapas et al., 2013). In addition, the connection with nonverbal reasoning is consistent with the need for higher-level reading comprehension skills (Fuchs et al., 2012). Language comprehension involved vocabulary and grammar (conform Kintsch & Rawson, 2005). Predictors for decoding were not found in the present control group, possibly because these measures were aimed at a lower reading level and were not sensitive enough for differences in decoding abilities at a higher level.

The structure of the path model we found for the ID group was similar to the control group, with some exceptions that are consistent with their lower reading level. Decoding was the most important predictor for reading comprehension, which is a common observation in early readers (Vellutino et al., 2007). Furthermore, decoding was connected to rapid naming and early literacy skills (consistent with studies in typical development and ID; Melby-Lervåg et al., 2012; Soltani &

Roslan, 2013). Vocabulary was only connected to reading comprehension through listening comprehension and was not connected to decoding (Perfetti, 2007). This is consistent with the reading profile of typically developing children who are in the early reading stages (Engen & Høien, 2002; Ouellette & Beers, 2010). In early readers, reading has not yet been incorporated in the language system as it is in more advanced readers, making decoding skill a prerequisite for reading comprehension rather than language comprehension skills (Vellutino et al., 2007). For older, more skilled readers with Down syndrome those strong relationships of reading comprehension with language comprehension and vocabulary have indeed been found (Nash & Heath, 2011; Roch et al., 2011). It is also important to note that in transparent orthographies, word decoding has been found to depend less on the oral vocabulary than in complex orthographies (Seymour et al., 2003). This might further explain why vocabulary was not yet a prominent predictor in the present group.

More remarkably, the current study found a strong direct relationship between early literacy skills and reading comprehension on top of decoding. Where in the study of Dessemontet and De Chambrier (2015) the relationship between early literacy skills and reading comprehension might have reflected word decoding skill, in the present study it is shown that the two are strongly related, even when decoding is taken into account. Early literacy skills may additionally reflect a number of cognitive skills that are crucial in language comprehension, such as working memory and metacognitive processes (Engen & Høien 2002). In addition, temporal processing was directly related to reading comprehension. This signals that reading comprehension problems may at least partly be attributed to difficulties in the processing of serial, phonemic information streams. Restrictions in temporal processing can influence both word decoding and language processing (Huss et al., 2011; Malenfant et al., 2012), which may result in poor reading comprehension (Hoover & Gough, 1990). Temporal processing may also affect reading comprehension through the development of reading prosody (Miller & Schwanenflugel, 2008).

The present results show that children with mild ID follow the same reading path as typically developing children, in line with the simple view of reading (Hoover & Gough, 1990). As such, a similar course could be followed in reading education for this group, albeit at a slower pace and with an tailored approach where necessary. The important additional role of early literacy skills in our model signifies that reading education for individuals with ID should pay much attention to phonological and phonemic awareness and should continuously provide explicit instruction in speech-sound awareness and letter-sound awareness (Barker et al., 2013; Channell et al., 2013; Soltani & Roslan, 2013). Temporal processing was directly related to the process of reading comprehension, on top of linguistic predictors. This emphasizes the fundamental importance of cognitive skills for gaining literacy. It is important to carefully monitor and support literacy-related cognitive skills throughout primary school. Due to impairments in information processing, reasoning, and working memory, fewer resources are available for children with ID to support the word reading process, as well as the deduction of meaning from a text. Children with ID can learn to partly compensate for the disadvantage caused by their cognitive limitations. Intervention studies have shown that literacy in persons with mild to moderate ID improve substantially when comprehensive explicit instruction on reading skills are provided over a longer period of time (Allor et al., 2014; Finnegan, 2012). Development in the linguistic domain can be supported by using visual aids while engaging in meaningful learning (e.g. Van der Schuit, Segers, Van Balkom, & Verhoeven, 2011). It may also be beneficial to address the development of underlying cognitive skills in reading, such as pattern recognition, visual matching, reasoning, and working memory.

Several limitations apply to the present study. First, the present group was heterogeneous with regard to etiologies. A wide spectrum of syndromes and disorders was represented in our group, as was intended because we were primarily interested in the effect of an intellectual disability in general. However, there were several disorders that were seen on a regular basis, such as ADHD and autism. In a similar vein, large differences in reading level were visible within the ID group. Although this provides insight in the general reading patterns of children with ID, caution is necessary when applying these results to individuals. Second, the present control group only allowed for a direct comparison of test scores between the two groups of similar instructional age. Due to a large discrepancy in reading level, a direct comparison of the predictors for reading comprehension between groups was not meaningful. Ideally a second control group might have been included in the study, matched on literacy level. However, the existing literature on predictors for reading development in children with typical development is extensive, which allows for a confident interpretation of the present results in the light of the existing theoretical framework. Finally, the present study assessed all measures at one point in time, and as such is correlational and not causal. Although earlier research supports causal interpretation of these relationships, a longitudinal study must provide information about actual prediction and the role of any underlying factors in the development of literacy.

Based on the limitations of the present study, several suggestions for further research can be made. First, a larger number of participants would provide statistical power that allows distinguishing between participants of different etiologies and between good and poor decoders within the ID population. Much is still unknown about the additional effect of specific syndromes on top of intellectual disability. Second, given the large differences in word decoding and reading comprehension skill within the ID group, it is recommended that future studies include a reading level control group to enable direct comparison of the relationships between specific variables. Third, given the relative transparency of the Dutch orthography it will be necessary to investigate how the present results generalize to other languages. Finally, a longitudinal approach is necessary to verify the predictive value of the present model and the role of any underlying factors in the development of literacy in children with mild ID. Longitudinal data through the higher educational grades will also provide insight in further stages of literacy development for children with ID. Previous research (Seigneuric & Ehrlich, 2005) suggested a more pronounced influence of cognitive factors once basic word decoding is attained. When reading fluency increases, the contribution of language comprehension to reading comprehension may also increase, similar to typical reading development (Nash & Heath, 2011; Roch et al., 2011).

To conclude, children with ID lag behind on cognitive and linguistic skills that are related to reading achievement. The level of literacy varies greatly among children with ID of similar age. In general, the group showed a reading profile that was similar to typically developing children in first grade. For reading comprehension, both word decoding and language comprehension were identified as predictors in the path model, meaning that the simple view of reading also applies to individuals with ID, with an additional impact of early literacy skills and temporal processing. The strong relationship between reading comprehension and early literacy skills emphasizes the need to continue the training of phonological awareness and letter-sound matching throughout the reading education of individuals with ID. The present study makes it also clear that support of the cognitive aspects of language processing in children with ID can be recommended.

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4.

Cognitive Constraints on the Simple View of Reading: A Longitudinal Study in Children with Intellectual Disabilities

This paper is based on:
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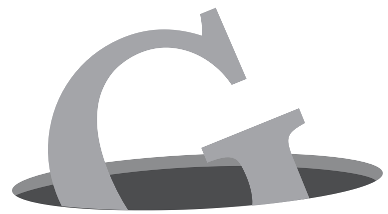

Abstract

The aim of the present longitudinal study was to verify 1) whether the simple view of reading applies when its cognitive basis is compromised, and 2) how limitations in cognitive and linguistic skills affect reading comprehension. The study included 78 children with mild intellectual disabilities (average IQ 60.45, age 121 months) in a three-year longitudinal design. The children were assessed for level of reading comprehension, decoding, listening comprehension, and underlying linguistic predictors (early literacy skills, vocabulary, grammar comprehension) and cognitive predictors (rapid naming, pseudoword repetition, nonverbal reasoning, temporal processing, and working memory). Results indicate that the simple view of reading applies in children with intellectual disabilities. Early literacy skills and nonverbal reasoning emerged as additional predictors for reading comprehension in this group. The outcome implies that cognitive disorders affect reading comprehension through limitations in both cognitive and linguistic skills.

Impairments in cognitive abilities severely affect literacy development and, in consequence, reading comprehension (Lemons et al., 2013). Reading comprehension is typically achieved by a combination of decoding skill and listening comprehension (Hoover & Gough, 1990; Language and Reading Research Consortium, 2015), supported by cognitive skills that facilitate attribution of meaning. However, it is not established what predicts reading comprehension when general cognition is constrained. The present study uses a longitudinal design to investigate how the simple view of reading applies in children with intellectual disabilities.

Longitudinal studies have shown that literacy-related skills early in life are predictive for reading achievement later on (Catts, Herrera, Nielsen, & Bridges, 2015). The main predictors for decoding are early literacy skills (letter knowledge and phonological awareness), rapid naming (Moll et al., 2014, Muter, Hulme, Snowling, & Stevenson, 2004), and in some cases pseudoword repetition (Perez, Maierus, & Poncelet, 2012). In addition, vocabulary assists word recognition (Muter et al., 2004) and temporal processing supports the perception of speech and analyzing the structure of speech (Walker, Hall, Klein, & Philips , 2006), which is a prerequisite for developing phonological awareness. Reading comprehension is predicted by prior decoding and listening comprehension skills such as vocabulary and grammatical knowledge (Muter et al., 2004), but also by early literacy skills in the first grade (Catts et al., 2015; Fuchs, Compton, Fuchs, Bryant Hamlett, & Lambert, 2012; Melby-Lervåg, Lyster, & Hulme, 2012). The level of reading comprehension can further be influenced by differences in temporal processing and attention regulation (Kim, 2016; McVay & Kane, 2012), as well as working memory and reasoning ability (Oakhill & Cain, 2012; Seigneuric & Ehrlich, 2005). Consequently, when cognition fails it is likely that literacy attainment will be affected as well.

Children with intellectual disabilities have limited reasoning and information processing capacity (Goharpey, Crewter, & Crewter, 2013; Schuchardt, Gebhardt, & Mäehler, 2010), which affects their literacy acquisition (Lemons et al., 2013). Knowledge about the underlying mechanisms of reading comprehension in children with intellectual disabilities is still fragmented. Predictors for decoding match the typical pattern (Soltani & Roslan, 2013; Wise, Sevcik, Romski, & Morris, 2010). However, the simple view of reading and its underlying predictors have not yet been established in a generic group of children with intellectual disabilities, in a longitudinal design. In adolescents with Down syndrome who were skillful decoders, listening comprehension was the main predictor for reading comprehension in a longitudinal study (Roch, Florit, & Levorato, 2011) and in a single trial study (Nash & Heath, 2011). A longitudinal study in children with mixed-etiology intellectual disabilities found that early literacy skills predicted reading comprehension, but decoding was not included as a predictor in the analysis (Dessemontet & De Chambrier, 2015). Decoding was found to be the main predictor for reading comprehension in children with mixed-etiology intellectual disabilities with basic decoding skills, within one timepoint (Van Wingerden, Segers, Van Balkom, & Verhoeven, 2014). One study focused on cognitive predictors for reading comprehension in late adolescents with velo-cardio-facial syndrome and borderline intellectual functioning. For this specific group, reading comprehension was predicted by decoding, attention, self-monitoring, and working memory (Antschel, Hier, Fremont, Farone, & Kates, 2014). In sum, predictors for reading comprehension seem consistent with the simple view of reading, but there is much variation in the setup of the few available studies, and most studies focus only on linguistic aspects of literacy.

The present paper explored how cognitive constraints influence the common theoretical framework for reading comprehension (Catts et al., 2015; Muter et al., 2004), in particular the simple view of reading (Hoover & Gough, 1990), in a longitudinal design. We explored whether cognitive abilities are only fundamental of the linguistic skills that lead to reading comprehension, or that cognitive factors also directly predict reading comprehension levels in children with intellectual disabilities. To this end we conducted a three-year longitudinal study, of which the data from Wave 1 have been reported in Chapter 3. Here we report the data from Waves 1, 2 and 3.

Method

Participants

The study was conducted in the Netherlands, in 20 schools for special education. Inclusion criteria were an IQ between 50 – 80, age between 108 and 138 months at Wave 1, normal hearing, eyesight that was normal or corrected to normal, and a monolingual Dutch-speaking home environment. There were no requirements with regard to reading level. Participants were tested at three time points, one year apart. At Wave 1, 81 participants (29 girls, 52 boys; IQ 50 – 80, *M* = 60.38, *SD* = 7.20) participated, mean age 121 months (*SD* = 5.83). At Wave 3, 78 participants (94%) were still included (28 girls, 48 boys; IQ 50 – 80, *M* = 60.45, *SD* = 7.49), mean age 145 months (*SD* = 5.80). Two participants dropped out due to a change of schools, one because of health reasons.

Measures

Participants completed several tasks that measured cognitive and linguistic skills related to reading comprehension. Unless specified otherwise, the reported test score is the number of correct responses on a task.

Wave 1

Early literacy skills were measured using a computer test (Vloedgraven, Keuning, & Verhoeven, 2009). Four competences were assessed: rhyme, synthesis, deletion, and letter knowledge. In three tests, one out of three pictures had to be selected. For *rhyme*, participants selected the picture representing a word rhyming with an auditory stimulus. In the *synthesis* task, participants heard a string of phonemes that spelled the target word. In the *deletion* task, participants heard a word of which they had to delete a certain phoneme, which resulted in a new word. In the *letter knowledge* task, participants had to select one out of four letters or digraphs corresponding to an auditory stimulus. Cronbach's alpha = .78 (Egberink, Vermeulen, & Frima, 2014).

Vocabulary was tested using the Dutch version of the PPVT-III-NL (Schlichting, 2005). During each trial, the participant chose one out of four pictures that best represented a verbally presented word. Item difficulty increased until a ceiling level was reached. Lambda-2-coëfficient = .93 - .97.

Grammar comprehension was tested using a multiple choice task comprising aurally presented sentences that required understanding of meaning relations within each sentence (Verhoeven & Vermeer, 2001). Cronbach's alpha = .81 (Verhoeven & Vermeer, 2006).

Rapid naming included five different line drawings of familiar objects, repeated in random order along four columns of 30 items, presented on one page (Verhoeven, 2005). The participant named the pictures in the presented order, during one minute. Cronbach's alpha = 0.95.

Pseudoword repetition comprised aurally presented pseudowords, which the participant was asked to repeat (Verhoeven, 2005). Phonemic complexity of the words increased until a ceiling level was reached. Cronbach's alpha = .94.

Working memory was tested using a Digit Span test (WISC-III-NL; Kort et al., 2005). Participants repeated strings of digits, increasing in length from 2 to 9 items, until a ceiling level was reached. First the digits were repeated in the same order, later the order had to be reversed. Cronbach's alpha = .64.

Temporal processing was measured using the Rhythm Test (Van Uden, 1983). Participants repeated rhythms of increasing complexity by tapping on the table with a pencil. They repeated each rhythm twice immediately after demonstration, and then five times without further demonstration. The accuracy of the repetition was rated. The test continued until all a ceiling level was reached.

Nonverbal reasoning was tested using Raven's Coloured Progressive Matrices (Raven, 1958). Participants chose one out of six puzzle pieces that best completed a larger visual pattern. Split-half reliability = .82 - .87 (Van Bon, 1986).

Wave 2

Decoding was measured by word decoding, and pseudoword decoding. Word decoding was assessed with three word lists, each containing 120 - 150 words of different orthographic complexity. Participants read as many words as possible in 1 minute from each card. The total number of correct responses was counted (Krom & Jongen, 2009). Cronbach's alpha = .86 - .94 (Krom, Jongen, Verhelst, Kamphuis, & Kleintjes, 2010). Pseudoword decoding was a similar task to word decoding, except that lists of pseudowords were used. Participants were given two minutes for each list (Verhoeven, 2005). Cronbach's alpha > .85.

Listening comprehension was tested by reading three short stories to the children (CELF 4-NL; Kort, Compaan, Schittekatte, & Dekker., 2008). After each story, five open-ended questions were asked that either directly referred to the story content, or required additional reasoning. Cronbach's alpha = .71 - .74 (Egberink et al., 2014).

Wave 3

Reading comprehension was measured by eight short stories with three multiple-choice questions, appropriate for the first grade of regular education. Four stories focused on using anaphora (Verhoeven 1992) and four stories focused on making inferences (Aarnoutse 1997). Cronbach's alpha = .85 for the anaphora task and .75 for the inferences task.

Procedure

The schools selected participants fitting the inclusion criteria and obtained active parental consent before the testing started. On all three time points, participants were tested during school hours in a separate room, by the first author and five undergraduate students of Educational Science who had received extensive training to follow standardized instructions.. Testing took 120 minutes at Wave 1, 30 minutes at Wave 2, and 60 minutes at Wave 3. Test assessment was always split up in 30-minute sessions.

Data Analysis

Participants who only participated at Wave 1 were excluded from the analyses. Missing scores (< .01% in total), were estimated based on previous or later scores on similar tasks from the larger longitudinal data set. We calculated z-scores for all scores to adjust for differences in scale and distribution between variables.

To reduce the number of variables, we created composite scores based on a confirmatory factor analysis, by adding the z-scores of related measures: reading comprehension consisted of anaphoric and inferential reading comprehension, early literacy skills consisted of rhyme, synthesis, deletion and letter knowledge, and decoding consisted of word decoding and pseudoword decoding. The composite scores were then treated as measured variables in further analysis. Skewness and kurtosis were adequate for all variables except for temporal processing, due to three participants with very high scores. Stepwise multiple regression analysis was used to identify the main predictors underlying reading comprehension, decoding, and language comprehension. Structural equation modeling analyses in LISREL 8.80 was used to further evaluate the relationships between cognitive and linguistic predictors and the outcome measure. The model fit was evaluated according to six indices: the χ^2:df ratio should be lower than 2:1, χ^2 significance should exceed .05, the NNFI, CFI, and GFI should exceed .90, and the RMSEA should be below .05 (Baumgartner & Homburg, 1996; Ping, 2004).

Results

The means and standard deviations for all scores are displayed in Table 1.

Table 1.

Descriptive Statistics for All Timepoints.

	Max. score	Exp. score	Wave 1 *M*	Wave 1 *SD*	Wave 2 *M*	Wave 2 *SD*	Wave 3 *M*	Wave 3 *SD*
Reading comprehension								
Anaphora	12	12					7.50	3.99
Inferences	12	12					6.67	3.15
Decoding								
Word decoding (3 mins)	-	292			88.46	82.92		
Pseudoword decoding (6 mins)	-	372			97.24	107.08		
Listening comprehension	15	13			6.36	4.08		
Early literacy skills								
Rhyme	15	15	11.09	3.24				
Synthesis	15	15	12.67	2.72				
Deletion	15	15	6.73	3.30				
Letter knowledge	34	34	31.05	5.49				
Vocabulary	-	125	88.14	15.59				
Grammar	42	42	30.04	6.53				
Rapid naming	120	61	45.35	14.06				
Pseudoword repetition	40	28	12.58	9.24				
Working memory	30	13	7.14	2.35				
Rhythm	-	115	27.38	36.76				
Nonverbal reasoning	36	31	19.92	6.11				

Note. Max. score = Maximum score on test, if applicable. Exp. score = expected score on test, based on mean age of participants.

First, we tested whether the simple view of reading applied. A multiple regression analysis was performed with reading comprehension (Wave 3) as the criterion variable, and decoding and listening comprehension (Wave 2) as predictors. Results showed that both decoding (β = .57, $p < .001$) and listening comprehension (β = .41, $p < .001$), were significant predictors of reading comprehension, $R^2_{adj.} = .56$, $F(2,75) = 50.36$, $p < .001$.

Second, we looked for additional cognitive or linguistic predictors (Wave 1) for reading comprehension. Table 2 presents a summary of all correlations between predictors at Wave 1 and criterion variables at Wave 2 and 3. Because of the large number of variables, we adopted a significance level of .01 for this analysis. Still, most predictor variables were moderately or strongly correlated at Wave 1, except pseudoword repetition and rapid naming, and rhythm and rapid naming. Correlations with outcome measures also reached significance in nearly all cases, except between temporal processing and listening comprehension.

Table 2.

Correlations Between Predictor Variables at Wave 1, and Outcome Variables at Wave 2 and 3.

	Wave	1	2	3	4	5	6	7	8	9	10
1. Reading comprehension	3	-									
2. Decoding	2	.64**	-								
3. Listening comprehension	2	.51**	.18	-							
4. Early literacy skills	1	.77**	.54**	.48**	-						
5. Vocabulary	1	.57**	.33*	.65**	.58**	-					
6. Grammar	1	.72**	.41**	.61**	.68**	.74**	-				
7. Rapid naming	1	.54**	.54**	.37*	.52**	.36*	.42**	-			
8. Pseudoword repetition	1	.53**	.40**	.28	.50**	.50**	.40**	.19	-		
9. Working memory	1	.66**	.52**	.39**	.68**	.56**	.60**	.49**	.48**	-	
10. Temporal processing	1	.36*	.31*	.10	.48**	.31*	.33*	.21	.52**	.46**	-
11. Nonverbal reasoning	1	.60**	.28	.35	.55**	.46**	.61**	.39**	.33*	.54**	.43**

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

Before building up the path model of predictors for reading comprehension, stepwise multiple logistic regression was used to identify the main cognitive and linguistic variables at Wave 1 predicting reading comprehension at Wave 3, and decoding and listening comprehension at Wave 2. In the first step of each analysis, cognitive predictor measures (rapid naming, pseudoword repetition, nonverbal reasoning, temporal processing, and working memory) were entered using the forward selection method. In the second step, linguistic predictors (early literacy skills, grammar comprehension, and vocabulary) were added with the same forward selection method. Reading comprehension at Wave 3 was related to rapid naming (β = .26, $p = .003$), pseudoword repetition (β = .26, $p = .002$), working memory (β = .26, $p = .011$), and nonverbal reasoning (β = .27, $p = .003$) in step 1, $R^2_{adj.} = .60$, $F(4,73) = 29.40$, $p < .001$. When adding linguistic predictors in step 2, the significant predictors for reading comprehension were rapid naming (β = .16, p = .040), pseudoword repetition (β = .16, p = .035), early literacy skills (β = .32, p = .004), and grammar comprehension (β = .25, p = .010), $R^2_{adj.}$ = .69, ΔR^2 = .10, F(6,71) = 29.85, p < .001. The other predictors from step 1 were no longer significant. Decoding at Wave 2 was predicted by rapid naming (β = .48, p < .001) and pseudoword repetition (β = .31, p = .001) in step 1 ($R^2_{adj.}$ = .37, F(2,75) = 23.57, p < .001), and by rapid naming (β = .37, p = .001), pseudoword repetition (β = .21, p = .043), and early literacy skills (β = .24, p = .047) in step 2 ($R^2_{adj.}$ = .39, ΔR^2 = .03, F(3,74) = 17.72, p < .001). Listening comprehension at Wave 2 was predicted by working memory (β = .39, p < .001) in step 1 ($R^2_{adj.}$ = .14, F(1,76) = 13.50, p < .001), and by vocabulary (β = .44, p = .001) and grammar (β = .31, p = .025) in step 2 ($R^2_{adj.}$ = .44, ΔR^2 = .31, F(3,74) = 20.99, p < .001).

With respect to the path model, structural equation modeling was used to construct a framework of the main predictors for reading comprehension in children with intellectual disabilities. All predictor measures identified by the regression analyses were added to this model. Non-significant paths were removed. In the final model (Figure 1) reading comprehension was directly predicted by decoding and listening comprehension (Wave 2), as well as early literacy skills and nonverbal reasoning (Wave 1; R^2 = .75). Decoding, in turn, was predicted by early literacy skills and rapid naming (Wave 1; R^2 = .42), and listening comprehension was predicted by vocabulary and grammar (R^2 = .45). The fit of the final model was excellent (χ^2(10) = 11.17, p = .420, NNFI = 0.99, CFI = 1.00, GFI = 0.97, RMSEA = .018).

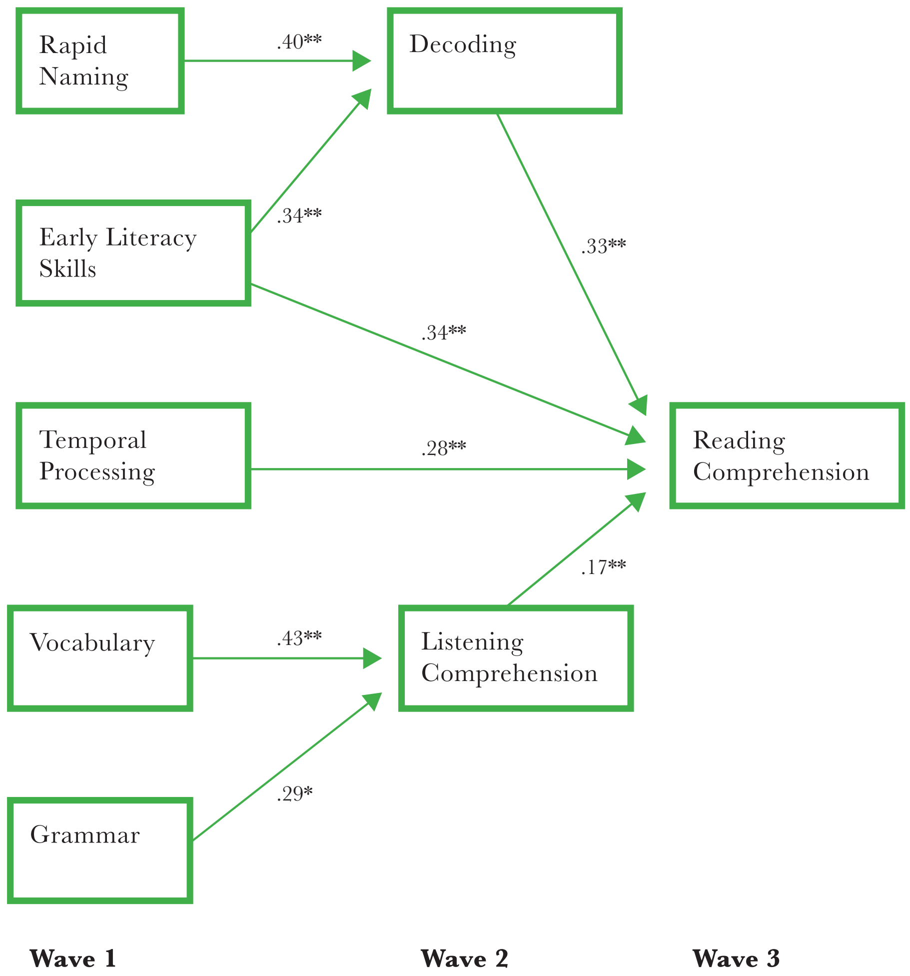

Figure 1.
Final path diagram to show the relations between cognitive and linguistic predictors at Wave 1, decoding and listening comprehension at Wave 2, and reading comprehension at Wave 3. Standardized beta weights are shown. All shown paths are significant.

Discussion and conclusion

The present study examined how cognitive constraints may affect existing models of reading comprehension. The findings support the general validity of the simple view of reading. The simple view of reading (Hoover & Gough, 1990) was found as the main predictive model for reading comprehension, even in children who experience cognitive constraints due to intellectual disabilities. The typical linguistic predictors for decoding and listening comprehension were also identified in the present model (Catts et al., 2015; Muter et al., 2004), in a pattern that is similar to early readers (see also Chapter 3). The long-term predictors for reading comprehension are also very similar to a study by Fuchs et al. (2012), who identified Grade 1 decoding, language comprehension, phonological processing and nonverbal reasoning as significant predictors for Grade 5 reading comprehension in typically developing poor readers.

The direct relationship between reading comprehension and nonverbal reasoning abilities implies that reading comprehension requires additional cognitive processing skills for pattern recognition and problem solving. Working memory was related to reading comprehension and listening comprehension, but only when linguistic skills were not taken into account. Still, the combination of cognitive predictors in Step 1 explained almost the same percentage of variation in reading comprehension as *with* inclusion of linguistic predictors in the model. For decoding a similar pattern was observed. This signals that reading comprehension is directly influenced by limitations in cognition, as well as through limitations in the linguistic domain. Moreover, early literacy skills were directly predictive for reading comprehension even when decoding was taken into account. This has previously been observed in early readers with typical development by Engen and Høien (2002), who pointed out that early literacy skills may partly reflect related cognitive skills such as working memory and metacognitive awareness. A similar point seems to be made by Fuchs et al. (2012), who emphasize the importance of cognitive characteristics for reading development.

It seems that for the present group early literacy skills are fundamental for decoding as well as reading comprehension, and a restraint at this lower level of literacy affects the higher level reading skills. The results highlight the importance of good early literacy skills for later reading comprehension levels, and also emphasize that cognitive skills should not be underestimated in literacy education. It is essential to identify a student’s strengths and weaknesses in the cognitive domain in order to strengthen any problem areas.

A comment can be made with regard to the sampling of the present study. All children with a certain level of IQ and age were included, regardless of etiologies. This resulted in a heterogeneous sample, which is reflected in the variation in literacy level of the participants. The diversity within the group, however, can be seen as a strength, as it gives a representative impression of the development of reading in children with intellectual disabilities, and not just that of a specific subgroup.

In conclusion, the simple view of reading continues to apply when cognition is constrained, with early literacy skills and nonverbal reasoning as additional predictors to the model. Cognitive constraints affect reading comprehension through linguistic predictors and through reasoning skills.

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Ch. 4

5.

Reading Comprehension in Children with an Intellectual Disability who are Deaf or Hard of Hearing

This paper is based on:
Van Wingerden, E., Segers, E., Van Balkom, H. & Verhoeven, L. (resubm). Reading Comprehension in Children with an Intellectual Disability who are Deaf or Hard of Hearing. Manuscript resubmitted for publication.

Abstract

This study aimed to gain insight in the literacy level of children with an intellectual disability who are deaf or hard of hearing (ID-DHH), and in the cognitive and linguistic factors that may impact their literacy attainment. Twenty-nine children with ID-DHH (age 102 – 166 months) were compared to 81children with an intellectual disability (ID; age 107 – 137 months) and 84 children with typical development (TD; age 99 – 126 months) on cognitive and linguistic predictors of literacy. Outcome measures were phonological awareness (rhyme and synthesis), letter knowledge, decoding, and reading comprehension. Children with ID-DHH performed severely below both reference groups and were mostly in a pre-literate stage. Phonological awareness was weak overall, but letter knowledge was reasonable for most participants. Several older children had achieved a basic level of conventional literacy. Within-group comparison was used to differentiate between stronger and weaker readers in the ID-DHH group. Better readers within the ID-DHH group were mainly distinguished by a larger vocabulary and better decoding skills than weaker readers. Despite the low reading levels overall, the patterns of predictors in the ID-DHH group were consistent with existing literature on literacy development in children with TD and ID, and literacy development in children who are DHH.

Literacy learning is impaired in individuals with an intellectual disability (ID; Lemons et al., 2013), as well as in individuals who are deaf or hard of hearing (DHH; Lederberg, Schick & Spencer, 2012). The prevalence of hearing loss among individuals with mild to moderate ID is estimated between 21% and 51%, depending on the type of measure and criteria (Carvill, 2001; Meuwese-Jongejeugd et al., 2006). Despite this common co-occurrence, there are very few studies that examined reading in children with ID who are also DHH (ID-DHH). The present study aimed to describe the reading level of 29 children with ID-DHH, alongside their variations in hearing, cognitive skills, and linguistic abilities.

Typical reading development

For reading, it is necessary to connect written text to spoken language (Hoover & Gough, 1990). Children with a typical development (TD) learn to master this skill in the early grades of primary school. The development of word decoding skill is mainly predicted by the level of phonological awareness, letter knowledge, rapid naming, and verbal short-term memory (Melby-Lervåg, Lyster, & Hulme, 2012; Moll et al., 2014). When word decoding skill is sufficiently internalized, the emphasis shifts to reading comprehension. To achieve full literacy and comprehend larger texts, it is necessary to know word meanings to a deeper level (as is explained in the lexical quality hypothesis; Perfetti & Hart, 2002), as well as to comprehend the structure of the corresponding spoken language; its syntax and discourse (Hoover & Gough, 1990; Vellutino, Tunmer, Jaccard, & Chen, 2007). Several cognitive skills are also involved in reading comprehension. Auditory perception and temporal processing are necessary for speech perception, grasping the order of phonemes and words, and detecting the prosodic patterns in spoken language (Cumming, Wilson, Leong, Colling, & Goswami, 2015; Malenfant, Grondin, Boivin, Forget-Dubois, Robaey, & Dionne, 2012). Working memory and reasoning ability are involved in text integration and constructing a meaningful message from the text (Stevenson, Bergwerff, Heiser, & Resing, 2014).

Reading development in children with ID

In children with ID, learning to read is hampered by limitations in cognitive capacity. Still, basic literacy is achievable for most children with ID if they are provided with regular, explicit instruction (Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2014; Lundberg & Reichenberg, 2011) supplemented with assistive or Augmentative and Alternative Communication (AAC) tools (Kliewer, 2008). The final level of literacy in children with ID is distributed over a wide range, from an emergent literacy level to basic reading comprehension (Jones, Long, & Finlay, 2006; Lemons et al., 2013). Predictors for word decoding are phonological awareness, letter-sound knowledge, rapid naming, and in some cases verbal short-term memory, matching the precursors that are observed in children with TD (Barker, Sevcik, Morris, & Romski, 2013; Channell, Loveall, & Conners, 2013; Dessemontet & De Chambrier, 2015). Reading comprehension in children with ID is strongly influenced by difficulties with word decoding (Channell et al., 2013). When word recognition is sufficiently automatized, reading comprehension becomes more strongly related to language comprehension skills, similar to children with typical development (Nash & Heath, 2011; Roch, Florit, & Levorato, 2011). However, individuals with ID generally have limited auditory information processing, as well as limited working memory and reasoning ability (Dessemontet & De Chambrier, 2013; Goharpey, Crewter, & Crewter, 2013; Schuchardt, Gebhardt, & Mäehler, 2010). Disorders in meaning attribution evoke difficulties in language comprehension, which is seen as a second barrier for reading comprehension in individuals with ID (Tavares, Fajardo, Ávila, Salmerón, & Ferrer, 2014).

Reading development in children who are DHH

The literacy development of children who are DHH is affected by limitations in the access to oral language (Nelson & Crumpton, 2015). Hearing technology such as hearing aids or cochlear implants support speech perception, although the auditory signals from this technology are not equivalent to normal hearing (Johnson & Goswami, 2010; Nelson & Crumpton, 2015). On average, word identification levels in children who are DHH are close to age-appropriate levels (Wauters, Van Bon, & Tellings, 2006), although a delay may be observed in older children (Harris & Terlektsi, 2010). The level of reading comprehension often stays below average, even in children with cochlear implants (Kyle & Cain, 2015; Marschark, Rhoten, & Fabich, 2007; Wauters et al., 2006).

Attainment of word decoding by means of pure grapheme-phoneme mapping is problematic for most children who are DHH; their reading vocabulary must partly be built by alternative strategies to achieve word-to-concept mapping (Perfetti & Sandak, 2000). Vocabulary is therefore major factor in the reading development of individuals who are DHH (Kyle & Harris, 2010; Mayberry, Del Giudice, & Lieberman, 2011). Connections between word decoding and phonological awareness have been evidenced as well, although the nature of this relation is still unclear (Bélanger, Baum, & Mayberry, 2012; Goldberg & Lederberg, 2015; Mayberry et al., 2011). Reading comprehension in individuals who are DHH is complicated by a discrepancy in rules and grammar between sign language and written language (Lederberg et al., 2012). The literacy attainment of individuals who are DHH is therefore influenced by variations in access to oral language such as the degree of

hearing loss, the use of different hearing devices, and speechreading ability (Johnson & Goswami, 2010; Knoors & Marschark, 2014; Kyle & Harris, 2010). Children who prefer communication through sign language are found to have lower reading comprehension skills than children who are DHH who use oral language (Kyle & Harris, 2010), although strong signing skills are also facilitative for better reading comprehension (Andrew, Hoshooley, & Joanisse, 2014; Cummins, 1981). In order to make use of all linguistic input in different modalities, cognitive processes such as attention, working memory, abstract reasoning, and executive functions, may be especially relevant for reading comprehension in individuals who are DHH (Daza, Phillips-Silver, Del Mar Ruiz-Cuadra, & López-López, 2014).

Reading in children with ID-DHH

Children with ID-DHH encounter barriers from both domains. An ID hinders the learning process in general due to limited cognitive capacity and a hearing loss hinders reading development through limited access to the oral language that the writing represents. In many developmental domains, including language development, these disabilities will mutually enforce each other; a deficit in one domain will hinder coping mechanisms to compensate for losses in the other domain, which can lead to diagnostic ‘overshadowing’ and misdiagnosis (Carvill, 2001; Meuwese-Jongejeugd et al., 2006). For example, learning difficulties due to ID may lead to a restricted sign vocabulary, or complicate the interpretation of acoustic signals that are provided by hearing technology. So far, research on individuals with ID-DHH has been scarce and consisted mainly of prevalence estimations (e.g., Meuwese-Jongejeugd et al., 2006; Timehin & Timehin, 2004). A case study was reported on the generalization of signing skills of a girl with hearing loss and moderate ID (Smeets & Striefel, 1976a,b), as well as a study on the use of sign language to facilitate communication (Hoffmeister & Farmer, 1972). More recent research investigated the effect of cochlear implants on the auditory perception and language development of children with ID-DHH. The reported effects of cochlear implants are substantial in children with mild ID, but small in children with moderate or severe ID (Daneshi & Hassanzadeh, 2007; Edwards, 2007). The main predictor for progress in language skills after implantation appears to be nonverbal cognitive skills rather than implant experience or age at diagnosis (Meinzen-Derr, Wiley, Grether, & Choo, 2009). The only reference we found that mentioned children with ID-DHH in relation to reading, is a study on children who are DHH with cochlear implants by Johnson and Goswami (2010). Six out of 78 children in that study had an IQ between 70 and 80, which was classified as borderline intellectual functioning according to the *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text rev.; *DSM–IV-TR*; American Psychiatric Association, 2000). The authors reported that three of these children read within one standard deviation of the expected attainment for their age and this subgroup seemed to have a similar language development pattern to children who are DHH (Johnson & Goswami, 2010).

The present study

The above presented literature review makes it clear that there is a gap in the knowledge on literacy abilities in children with ID-DHH, and the factors that are associated with these literacy abilities. Numerous studies have shown that children with severe disabilities, who had been expected to never gain literacy, progressed greatly when provided with suitable opportunities and encouragement (Browder, Gibbs, Ahlgrim-Delzell, Courtade, Mraz, & Flowers, 2008; Kliewer, 2008). The present study will be the first to regard children with ID-DHH as a group and use a typical research setting to gain insight in their common characteristics, in view of improving the literacy education for this group. In the Dutch special education system, the approach to literacy learning for children with ID-DHH is not clearly defined. Reading education for children with ID supports regular methods with visual aids such as photographs, visual symbols, and iconic hand gestures for words and letters. In special education for children who are DHH, regular methods are supported with signs and speechreading. Education for children with ID-DHH is in many respects more similar to education for children with ID than to education for children who are DHH, which was our motivation for choosing a reference group of children with ID for this study.

The literacy abilities (decoding and reading comprehension) of children with ID-DHH were compared to children with ID of similar age and to typically developing children of similar age. We also included a number of cognitive and linguistic predictors that influence literacy achievement in individuals with ID or who are DHH. Cognitive precursor measures were nonverbal reasoning, rapid naming, auditory perception, memory span, and temporal processing. Linguistic precursor measures were early literacy skills (phonological awareness and letter knowledge) and general language skills (vocabulary, grammar comprehension, and listening comprehension).

The first goal of this study was to gain insight in the current level of word decoding, reading comprehension, and precursor measures in children with ID-DHH compared to children with ID and TD. Reading is known to be weak in children with ID-HH, but the general reading levels have never been inventoried. Second, we aimed to distinguish cognitive and linguistic markers within the group of children

with ID-DHH besides hearing status or IQ, that could discern between children with ID-DHH of higher and lower literacy levels, relative to their peers. The finding of such markers may help determine the main issues in literacy development for this group. Finally, we investigated whether traditional predictors for decoding and reading comprehension were also related to literacy attainment in children with ID-DHH. This might aid in determining the type of support that is necessary for children with ID-DHH to reach their optimal literacy level.

Method

Participants

The scores of 29 participants with ID-DHH were compared to the scores of two reference groups of similar age: children with ID and children with TD (as reported in Chapter 3). Active parental consent was requested through an information letter for participation. Schools provided basic student information to the researcher, only when parental consent was given for participation.

The group of 29 participants with ID-DHH (13 male, 16 female) attended schools in The Netherlands that specialize in children who have communication problems and ID. Age within this group ranged from 102 to 166 months (M = 132.38, SD = 18.41), IQ ranged from 50 to 81 (M = 63.55, SD = 6.97). Hearing loss ranged between 50 – 150 dB on the unaided left side (M = 82.67, SD = 25.50) and 40 – 150 on the unaided right side (M = 82.41, SD = 30.20). Following the criteria of the World Health Organization (2013), 10 participants met the child criteria for moderate hearing loss, 4 had severe hearing loss, and 15 met the criteria for profound hearing loss including deafness. Age of identification was nine months at the latest, when a statutory hearing test was taken by the child health center. Further diagnosis took place around 15 months or later. Children who used cochlear implants had generally received a one-sided implant at an approximate age of 5-10 years, which was done with the main intent to prompt alertness and arousal and in some cases to support speech communication. Their language of preference was Dutch, Sign-Supported Dutch (SSD) or Sign Language of The Netherlands (SLN). None of the participants had parents who were DHH. Teachers use SSD for communication within the classroom. See Table 1 for a summary of participant details. More elaborate information can be found in Appendix A.

The ID reference group consisted of 81 children (52 male, 29 female) from 20 schools for children with ID in The Netherlands. They had a mild to moderate ID of diverse etiologies (IQ between 50 – 80, *M* = 60.38, *SD* = 7.20) and were between 107 – 137 months of age (*M* = 121.27, *SD* = 5.83). They were all native Dutch speakers, had normal or corrected eyesight and no diagnosis of hearing problems. A reference group of peers with TD consisted of 84 third-grade students (35 girls, 49 boys) from five schools for regular education in The Netherlands. Their age ranged from 99 – 126 months (*M* = 108.02, *SD* = 5.77). They were all native Dutch speakers. Eyesight, hearing level and intelligence fell within the normal range, i.e. they had no diagnosis of disability in these domains.

Materials

The participants completed several tasks that measured early literacy and its predictors. The reliability of all tests is high or very high for a hearing population. The reported test scores are the number of correct responses on a task, unless indicated otherwise.

Reading comprehension In the reading comprehension task, the participants read eight short stories, each followed by three multiple choice questions. The texts were appropriate for the first grade of regular education and focused on explicit anaphoric relations within the text (Verhoeven, 1992) and implicit inferential meaning (Aarnoutse, 1997). In the ID-DHH group, teachers were asked to guide their students through the example story and let them complete the task independently. In the ID group, the reading tasks were part of the test sessions. The reading comprehension level of TD participants was determined by their schools at least at second grade level, based on the Dutch national student monitoring system (Cito, 2007).

Decoding in the ID-DHH group was measured in an adapted version of a standardized test (Word decoding test for first grade; Jongen, Krom, & Roumans, 2009) that was also used in Van Tilborg, Segers, Van Balkom, & Verhoeven (2014). The participants selected the correct word from five similar looking words and pseudowords, of which one corresponded to a picture. The words increased in difficulty from CVC words to CVCVC words and two-syllable words. The task consisted of 40 items, but was terminated after four consecutive errors. For the remaining items a chance-level performance was estimated, which was added to the number of correct items.

Phonological awareness was measured by two computer tasks of 15 items each (Vloedgraven, Keuning, & Verhoeven, 2009). In the *Rhyme* task, participants selected one out of three pictures that corresponded to a spoken word. The three options were named, which was followed by 'what rhymes with…?'. In the *Synthesis* task, participants selected one out of three pictures that corresponded to the word that was formed by a string of 3 – 5 phonemes. The three options were named before the phoneme string was recited.

Letter knowledge The computer task for letter knowledge (Vloedgraven et al., 2009) included all 34 frequent letters and digraphs in the Dutch language. In each trial, the participant selected one out of four letters that corresponded to a phoneme. The target phoneme was named by the computer voice, e.g.: 'the *b* from *bear*'.

Vocabulary was measured using the PPVT-III-NL (Schlichting, 2005). Participants selected one out of four pictures that corresponded to a spoken word. Item difficulty increased after each set of 12 items. Ceiling level was reached when more than 9 errors were made within a set. The ID-DHH group started at set 4 (appropriate for children of 4;0 – 4;5 years), keeping in mind the translation to sign language of all items. The ID and TD control group started at set 6 (age 5;6 – 6;5)

Grammar comprehension was tested using the subtest Sentence comprehension from the Language Test for All Children (Verhoeven & Vermeer, 2001). The participants selected one out of three pictures that corresponded to an orally presented sentence. The task required understanding of explicit and implicit meaning relations within sentences. All 42 items were presented.

Listening comprehension was tested using three stories from the Dutch CELF-4-NL (Kort, Compaan, Schittekatte, & Dekker, 2008). The stories were read to the children, and each story was followed by five questions about the content and the meaning of the story, which the child answered in their chosen modality. The stories were between 67 and 115 words in length and were appropriate for typically developing children of 7 – 8 years.

Rapid naming was measured with a subtest of the Dutch Test for children with Specific Language Impairment (Verhoeven, 2005). Five pictures (duck, shoe, house, comb, and glasses) are repeated in 4 rows of 30 pictures on one page, in random order. The participants were instructed to name the pictures one by one, as rapidly and correctly as possible. For children with ID-DHH, speech and signs were both accepted, as long as the designation for each picture was consistent.

Auditory perception Words of increasing length and phonemic complexity were played from a laptop (Verhoeven, 2005). Participants were asked to repeat what they heard. For *word repetition* a maximum of 40 regular words were presented (increasing in length from one to five syllables, from 'lus' [loop] to 'vogelverschrikker' [scarecrow], for *pseudoword repetition* a maximum of 40 pseudowords were presented, with pseudoword complexity increasing in the same way. In both cases, the series was stopped after five incorrect answers in a row.

Memory Span Short-term memory span could be measured in two ways. The first was the standard test *Digit Span Forward* from the Dutch WISC-III-NL (Kort et al., 2005), where the participants repeated sequences of digits that increased in length from two digits up to nine digits, until they failed to repeat the series of a particular length twice. For participants with ID-DHH, the test was adapted to a signed *Letter Span Forward*, where each digit from the original test was replaced with a letter from the Dutch finger spelling alphabet. The letters were chosen so that the signs were least alike (C, V, M, G, B, L, S, T, K). In the ID-DHH group, both variants of the memory span task were administered if possible. Twenty-five participants with ID-DHH completed the Digit Span task and 26 completed the Letter Span task, of which 21 completed both Digit Span and Letter Span. For these 21 participants the two scores correlated strongly ($r = .84$, $p < .001$).

Temporal processing The rhythm test by Van Uden (1983) was used to test temporal processing. The participant repeated short rhythms of increasing complexity by tapping on the table with a pencil. An example was given twice by the experimenter, followed each time by an immediate repetition by the participant. Next, the participants repeated the same rhythm five more times. For non-hearing participants the example was given by tapping on the back of their hand. The test consisted of 15 test items in total, but was terminated after the direct imitation of a particular rhythm was incorrect twice.

Nonverbal reasoning Nonverbal reasoning skills were measured using Raven Colored Progressive Matrices (Raven, 1958). The test has 36 items, each requiring the participant to select one out of six visual patterns that best completes a larger pattern.

Procedure

The tests were administered separately in a quiet room within the schools. Test administration was divided over multiple test sessions that lasted a maximum of 30 minutes.

Table 1.

Demographic Information of Participants with ID-DHH. For Further Details on Individual Participants, see Appendix A.

	n
Age	
8 – 9	9
10-11	14
12-14	6
Gender	
Male	13
Female	16
Hearing loss better unaided ear	
40-60	9
60-80	4
>80	16
IQ	
71-81	1
61-70	19
50-60	9
Language of preference	
SSD	13
SLN	10
Speech	6
Hearing device	
CI	13
BTE	10
BAHA	3
None	3

Note. CI = Cochlear Implant, BTE = Behind The Ear hearing aid, BAHA = Bone Anchored Hearing Aid, SSD = Sign Supported Dutch, SLN = Sign Language of The Netherlands.

For the ID-DHH group, test administration took 120 minutes in total. Tests were administered by the first author and two undergraduate students in behavioural science. A teacher or teacher assistant accompanied the participant and translated the instructions in Sign Supported Dutch (SSD) or Sign Language of The Netherlands (SLN) if the participant preferred this over oral communication (see also Appendix A). If possible tests were adimistered orally or via SSD, because grammar and vocabulary would be more equal to the typical situation. Tasks were selected to enable nonverbal responses as much as possible, so that speech was not a requirement for participation. Table 2 sums up the number of students that required translation from oral language for all tasks that typically require a verbal response or stimulus presentation (memory span is mentioned above).

For the ID reference group, test administration took 120 minutes in total and was completed by the first author and two undergraduate students in educational science who had received extensive training for test administration. For the TD reference group the tests took 90 minutes because reading comprehension was assessed by the teacher. The remaining tests were administered by four undergraduate students in educational science who had been trained to follow the standardized test instructions.

Table 2.

Number of Students who Preferred to Receive Stimuli or Respond in Verbal Language, Sign Supported Dutch (SSD), Sign Language of The Netherlands (SLN), or Not in Either Modality (NA).

	Rhyme	Synthesis	Letter knowledge	Vocabulary	Grammar	Listening comprehension	Rapid naming (responses)
Verbal	15	16	18	17	12	5	11
SSD	4	6	7	7	9	7	13
SLN	6	5	4	5	7	13	5
NA	4	2	0	0	1	4	0

Data analysis

To compare test scores between groups, independent samples *t*-tests were used. To compare scores within the ID-DHH group, we performed nonparametric Mann-Whitney *U* tests and Spearman correlation due to sample size and unequal score distributions.

The ID-DHH group was divided in groups of high and low scorers on four stages of traditional literacy development: phonological awareness, letter knowledge, decoding, and reading comprehension. The division was made based on a cut-off point for chance level performance, taking into account a baseline score for guessing. The following formula was used: $\textit{cut-off point} = np + (n - np)p$, where n is the total number of items in the test and p is the chance of randomly selecting the correct answer on any item. The score for *phonological awareness* was a sum score of the rhyme task and the synthesis task. The total of 30 questions of 3 options resulted in a cut-off point at 16.7. *Letter knowledge* consisted of 34 items with 4 options, resulting in a cut-off point at 14.9. Because the *decoding* task included a termination rule, we assumed chance level performance on the remaining items for each participant and added 1/5 * the number of remaining items to the number of correct answers. Based on 40 items of each 5 options, the cut-off point was 14.4 for this task. Finally, for *reading comprehension* the cut-off point was 10.5, based on 24 questions of 4 options each.

To investigate mutual relationships between predictor variables and the outcome variables, Spearman rank correlations were used.

Results

Comparisons between reference groups

The test scores of the ID-DHH group were compared to those of the ID group and the TD group (Table 3). The ID-DHH group scored below the ID group, which in turn scored below the TD group on nearly all tasks (ID-DHH compared to ID, all *p's* < .01, *d* = 0.24 − 1.37; ID compared to TD all *p's* < .01, *d* = 1.32 − 5.32). The ID-DHH group was comparable to the ID group on IQ ($t(100) = 1.89$, $p = .061$, $d = 0.49$) and nonverbal reasoning ($t(108) = 0.47$, $p = .636$, $d = 0.20$).

Comparisons within the ID-DHH group

In order to identify cognitive or linguistic markers for children with ID-DHH who have stronger versus weaker literacy skills, differences between high and low scorers on four levels of literacy (phonological awareness, letter knowledge, word decoding, and reading comprehension) were calculated with a nonparametric Mann-Whitney *U* test (reported in Appendix B). The effect size estimate *r* was calculated according to Rosenthal (1991; $r = Z/\sqrt{N}$). A complete overview of the analyses can be found in Appendix B.

Regarding phonological awareness, of 25 participants who completed the test, 14 scored above chance level and 11 scored below chance level. There was a significant difference on the separate tasks between high and low scorers on both synthesis (*Mdn*high = 9, *Mdn*low = 6, $U = 26.5$, $p = .004$, $r = .56$) and rhyme (*Mdn*high = 12, *Mdn*low = 7, $U = 12.5$, $p < .001$, $r = .71$). Children who scored higher on phonological awareness had slightly lower IQ scores (*Mdn* = 62) than children with lower scores (*Mdn* = 67, $U = 36$, $p = .025$, $r = .45$), but were better at word repetition (*Mdn*high = 5, *Mdn*low = 0, $U = 32.5$, $p = .013$, $r = .54$).

For letter knowledge, 26 of 29 participants scored above chance level on this task (*Mdn* = 31), while only 3 participants scored below chance level (*Mdn* = 14, $U = 12.5$, $p < .001$, $r = .71$). No significant differences could be found between high and low scorers on other tasks.

Regarding decoding, 13 of 29 participants scored above chance level (*Mdn* = 30) and 16 scored below (*Mdn* = 10, $U = 0$, $p < .001$, $r = .85$). On the remaining tasks, only a marginally significant difference was found on age between participants with high scores (*Mdn* = 138 months) and participants with low scores (*Mdn* = 124.5 months, $U = 60$, $p = .056$, $r = .36$).

The reading comprehension task was administered in 15 cases. For the other 14 participants (7 scoring above chance level on decoding, *Mdn* = 21.60, and 7 scoring below chance level, *Mdn* = 10.20), their teachers refused to hand the assignment to their students, because they believed the task to be too difficult and as a result would frustrate the children too much. Of the 15 participants who completed the task, 5 had a score on reading comprehension above the cutoff point (*Mdn* = 14) and 10 scored below the cutoff point (*Mdn* = 6, $U = 0$, $p = .001$, $r = .80$). Participants who scored above chance level on reading comprehension had a larger vocabulary (*Mdn* = 97) than weaker comprehenders (*Mdn* = 68.5, $U = 6.5$, $p = .019$), and were better at decoding (*Mdn* = 34.4) than weaker comprehenders (*Mdn* = 10.3, $U = 3$, $p = .005$, $r = .70$).

Spearman correlations between precursor measures and reading outcome measures are displayed in Table 4. Phonological awareness was moderately related to word repetition ($rs(23) = .58$, $p = .003$), pseudoword repetition ($rs(23) = .54$, $p = .005$), and letter knowledge ($rs(23) = .43$, $p = .032$). Word decoding was moderately related to synthesis ($rs(25) = .40$, $p = .041$). Reading comprehension was moderately related to nonverbal reasoning ($rs(13) = .55$, $p = .032$), Rapid naming ($rs(13) = .52$, $p = .045$), letter span ($rs(13) = .56$, $p = .029$), vocabulary ($rs(13) = .62$, $p = .014$), listening comprehension ($rs(12) = .58$, $p = .032$) and strongly related to word decoding ($rs(13) = .77$, $p = .001$). Plots of the distribution of scores on decoding relative to traditional precursors (rapid naming, phonological awareness and letter knowledge) are displayed in Figure 1. The relationships between reading comprehension and traditional precursors (decoding, vocabulary and listening comprehension) can be found in Figure 2.

Table 3.

Scores and Comparisons for Children with an Intellectual Disability who are Deaf or Hard of Hearing (ID-DHH; N= 29), Intellectual Disability (ID; N = 81) and Typical Development (TD; N = 84).

			ID-DHH		ID		TD	
	n	Median	M (SD)	Range	*M (SD)*	Range	*M (SD)*	Range
Age	29	131	132.38 (18.41)	106-166	121.2 (5.81)	107-137	108.02 (5.77)	84-99
IQ	29	64	<u>63.55</u> <u>(6.97)</u>	50-81	<u>59.81</u> <u>(7.98)</u>	37-80		
Hearing loss left	27	80	82.67 (25.50)	50-150	NA		NA	
Hearing loss right	27	83	80.56 (30.20)	35-150	NA		NA	
Reading comp.	15	7	8.53 (5.25)	3-20	10.02 (6.48)	0-24	MAX	
Decoding	29	13	17.35 (10.26)	7-40			MAX	
Phonological awareness								
Rhyme	25	8	8.72 (3.10)	2-15	10.9 (3.24)	1-15	MAX	
Synthesis	27	7	7.67 (2.87)	4-13	12.43 (3.01)	4-15	MAX	
Letter knowledge	29	31	27.62 (7.17)	9-34	30.4 (6.80)	0-34	MAX	
Vocabulary	29	73	68.62 (29.30)	13-125	87.99 (16.04)	38-121	111.61 (8.76)	81-133
Grammar comp.	27	22	21.7 (4.91)	14-30	29.78 (6.80)	12-40	38.55 (2.44)	29-42
Listening comp.	25	6	5.36 (3.41)	0-12	7.43 (3.94)	0-15	11.55 (2.25)	6-15
Rapid naming	29	36	37.38 (16.32)	7-68	44.43 (14.86)	10-73	61.26 (9.58)	36-88
Auditory perception								
Word repetition	29	0	3.28 (6.60)	0-31			MAX	
Pseudoword repetition	29	0	1.17 (3.76)	0-20	12.25 (9.25)	0-34	28.05 (7.10)	5-39
Memory span								
Digit span	24	4	3.42 (1.84)	0-7	4.80 (1.66)	0-9	7.74 (1.68)	5-12
Letter span	26	3	2.65 (1.60)	0-6	NA		NA	
Temporal processing	29	4	8.41 (12.13)	0-51	27.04 (36.45)	0-172	114.85 (53.97)	4-210
Nonverbal reasoning	29	20	<u>21.03</u> <u>(6.71)</u>	7-34	<u>19.8</u> <u>(6.11)</u>	5-35	31.20 (3.43)	21-35

Note. Scores differ significantly between groups. <u>Underlined scores mean that *no* significant difference was found.</u> Comp. = comprehension. NA = not applicable for this group. MAX = scores estimated at ceiling level.

Table 4.

Spearman Correlations Between Reading Measures, and Cognitive and Linguistic Predictors.

	n	1	2	3	4	5	6	7	8	9	10	11	12	13	14
1. Reading comprehension	15	-													
2. Decoding	29	.77**	-												
3. Rhyme	25	-.03	.33	-											
4. Synthesis	27	.51	.40*	.38	-										
5. Letter knowledge	29	.35	.28	.23	.38	-									
6. Vocabulary	29	.62*	.32	.08	.31	.33	-								
7. Grammar	27	.42	-.04	.02	-.13	.13	.25	-							
8. Listening comprehension	25	.57*	.12	.06	.13	.26	.17	.54**	-						
9. Rapid naming	29	.52*	.14	.02	-.14	-.28	.05	.12	.02	-					
10. Word repetition	29	.26	.26	.40*	.48*	.15	.44*	.22	.09	-.10	-				
11. Pseudoword repetition	29	.11	.31	.42*	.41*	.05	.28	.22	-.01	-.20	.83**	-			
12. Digit span	24	.25	.21	.00	.12	.22	.28	.07	.19	.34	.25	.10	-		
13. Letter span	26	.56*	.34	.12	.27	.12	.38	.23	.47*	.41*	.34	.17	.84**	-	
14. Temporal processing	29	-.02	-.18	.10	-.04	-.26	.21	.16	.01	.29	.26	.08	.23	.26	-
15. Nonverbal reasoning	29	.55*	.37	.16	.19	-.05	.33	.04	-.10	.19	.25	.27	.20	.24	.08

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

Ch. 5

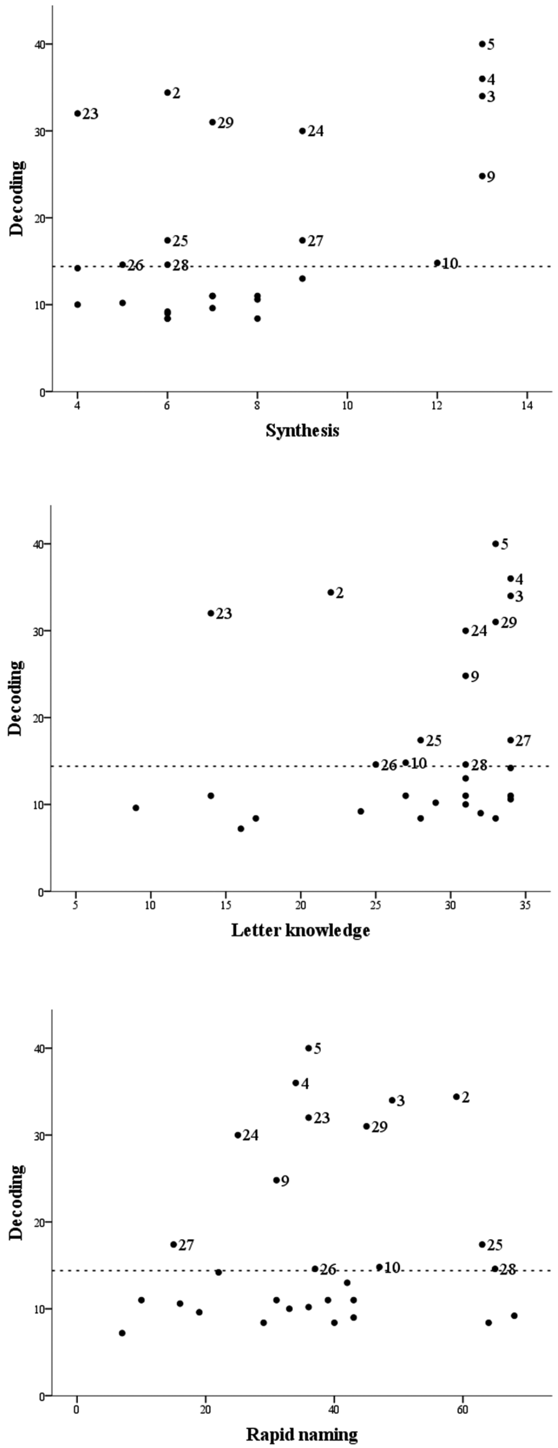

Figure 1.
Distribution of scores on synthesis, letter knowledge and rapid naming relative to decoding for participants with ID-DHH. Labels on each data point correspond to participant numbers in Appendix A. The dotted lines represent the boundary for chance level performance on the task.

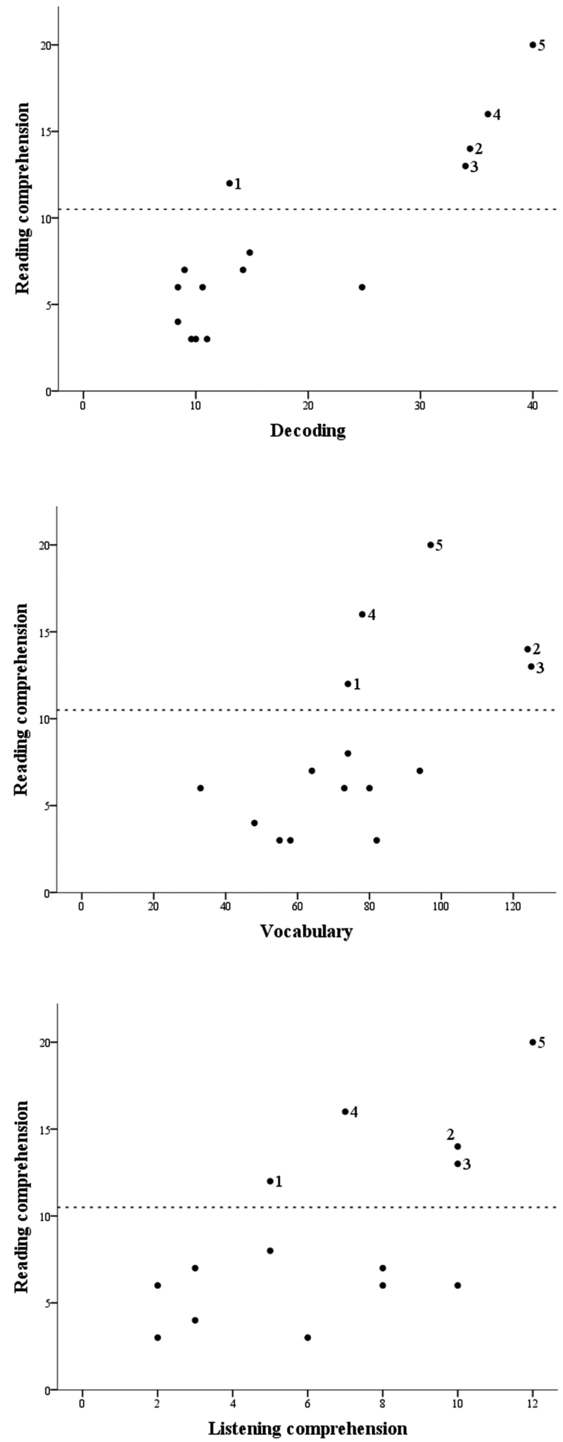

Figure 2.
Distribution of scores on decoding, vocabulary and listening comprehension relative to reading comprehension for participants with ID-DHH. Labels on each data point correspond to participant numbers in Appendix A. The dotted lines represent the boundary for chance level performance on the task.

Discussion

The present study was a first attempt to gain insight in the literacy of participants with ID-DHH. It was found that children with ID-DHH performed far below children with ID on literacy and reading-related skills. Their level of decoding and reading comprehension was still below First-grade level. Although most participants recognized a large number of letters, phonological awareness was still problematic for the majority of the group. Decoding, in the form of word recognition appeared to improve with age. Reading comprehension involved vocabulary in addition to decoding ability.

Group comparisons

Between groups, the comparison of children with ID-DHH to two control groups of similar age (ID and TD) reveals the severity of reading deficit in this group, even compared to a group of children with ID. The literacy level of the current ID-DHH group was also much lower than the average reading level that is reported in the literature for peers who are DHH (Wauters et al., 2006). Many of the participants with ID-DHH in the present study were still in a pre-literate phase and were not (yet) actively involved in word decoding at the end of primary school. It is striking to see that the ID-DHH group performed significantly below the ID group overall, despite some practical factors in the current test situation that may have worked to their advantage, as will be explained further below.

Within the ID-DHH group, nearly all participants scored high on letter knowledge, although many struggled with phonological awareness. This may indicate that phonological awareness does not necessarily precede the development of letter name knowledge in this specific group (see also Goldberg & Lederberg, 2015). Participants who were better at phonological awareness were somewhat better at repeating aurally presented words, which suggests an effect of hearing. They also seemed to have slightly smaller hearing loss than participants who were weak at phonological awareness, although this difference was not significant. However, there may have been an effect of hearing technology; relatively more participants had cochlear implants in the strong phonological awareness group (64 %) than in the weak phonological awareness group (36 %). The performance on decoding and reading comprehension were strongest in somewhat older participants, indicating that some children with ID-DHH can develop conventional literacy with time and patience at an older age. We found very few additional indicators for word decoding skill. The five participants with ID-DHH who were classified as strong reading comprehenders were better at decoding, had a larger vocabulary, and better listening comprehension than weak comprehenders, (Figure 1b; Hoover & Gough, 1990; Perfetti & Sandak, 2000). The strong relationship between decoding and reading comprehension is consistent with findings in early readers with TD (Vellutino et al., 2007). The relationship between vocabulary and reading comprehension is in line with literature on individuals who are DHH (Daza et al., 2014; Kyle & Harris, 2010) and the lexical quality hypothesis (Perfetti & Hart, 2002). The correlation analysis also yielded reasoning, memory and language comprehension as indicators for reading comprehension which are all related to cognitive processing and comprehension in both ID and DHH literature (Daza et al., 2014; Tavares et al., 2014)

Although the ID-DHH group performed severely below both the ID and TD reference groups, their pattern of predictors for literacy is in line with ID and DHH literature. The outcomes do not point to one dominant disability over the other. Vocabulary can be seen as the main linguistic predictor for reading comprehension, emphasizing that understanding written language starts with understanding signed or spoken language.

Individual differences in the ID-DHH group

The individual differences within the general pattern are visible in Figure 1 and 2. The scatter plot reveals that the relationship found between synthesis and decoding is mainly due to ceiling scores on synthesis by some of the good decoders, while others still had low scores (Figure 1). Note also Participant 2 and 23, who scored low on both synthesis and letter knowledge, but high on decoding. Both participants had a high degree of hearing loss and used no hearing device, which leads us to believe that they may read by means of orthographic word recognition, but had not yet extracted explicit phonological rules from these words (Bélanger et al., 2012; Goldberg & Lederberg, 2015). This hypothesis is supported by the additional high scores of participant 2 on vocabulary, language comprehension, and reading comprehension (Figure 2); his language skills and reasoning ability were sufficiently developed to comprehend the written text. Reversely, Participant 9 had good phonological awareness and letter knowledge, resulting in reasonable decoding skills (Figure 1), but her reading comprehension was hampered by weaker vocabulary and language comprehension skills.

The relationship between reading comprehension, decoding, vocabulary, and listening comprehension is reflected in the general score patterns (Figure 1b). However, exceptions can be found on an individual level: one good comprehender (Participant 1) was classified as a poor decoder. Closer inspection yields that her performance was around the group average or around threshold level on most tasks. Her score on decoding fell slightly below the established threshold, and her score on reading comprehension was slightly above the threshold. Her individual score pattern must therefore classified as mediocre, but consistent. This participant may continue to develop her literacy skills and further develop her decoding and comprehension skills.

While the general ID-DHH group adheres to well-known principles of reading comprehension, some individual differences emerge when looking at individual score patterns. Several participants were able to use their strongest linguistic skills (e.g., vocabulary, language comprehension) to compensate for weaker linguistic skills (e.g., synthesis, word decoding). Generally speaking, language skills seem decisive for the degree of compensation by an individual, for both decoding and reading comprehension.

Cautions and recommendations

Much caution is necessary when generalizing from this data. The group of participants was highly heterogeneous with regard to their additional diagnoses (see Appendix A). Schools did not always have recent or complete information available about hearing loss or IQ. Therefore we cannot report on individual characteristics in more detail, such as hearing loss after fit with hearing technology, or age of implantation. Concerning test results, there was high variability in score patterns between individuals and not all participants completed every test. Finally, in addition to factors that are measured by diagnostics, many other factors may influence these individual abilities, such as home environment, the specific educational track of a participant, and also the specific combination of personal traits (Knoors & Marschark, 2014; Lederberg et al., 2012).

In future research, some practical issues of testing should also be addressed. We decided to include teachers in the test sessions to facilitate communication with the experimenters and help the participants to feel comfortable in the unfamiliar situation. In practice, many different teachers accompanied the participants. Despite explicit instruction to only give a direct translation of instructions, some teachers were keen to help their students perform and avoid frustration. This resulted in repetition of questions when a wrong answer was given or rephrasing the instructions to make an implicit task more explicit. The same facilitative and protective approach of teachers was the reason why only half the ID-DHH group attempted the reading comprehension task. Both can be overcome by allowing only the researcher in the test room or by using one accompanist on each school who is completely briefed and instructed on the aims of the research and also familiar with the participants. A second issue to be addressed is that the current method of testing and test selection resulted in stimuli with relatively high iconicity when translated in sign language or sign supported Dutch. In many cases these iconic signs simplified task-performance (see Tolar, Lederberg, Gokhale, & Tomasello, 2007). For example, in the vocabulary task the sign for 'ball' is the shape of a ball. In the grammar task the original stimulus is 'the boy is pulled by the girl'. In SLN, the sentence is translated into 'boy in cart – girl pulling [the cart]', where the directionality of the pull is acted out by the translator. The same reasoning applies to a large portion of the Dutch sign alphabet being a direct visual representation of the letter shape, which may have boosted scores for letter knowledge in participants who requested signed stimuli. Future studies using a hearing reference group are advised to avoid the discrepancies that arise from translating speech-based tasks to visual signs, as described above. Where possible, tests should be used that are available and equivalent in both modalities. Finally, the present study contained several tasks that were initially played from a laptop for all groups (auditory perception, vocabulary, grammar, phonological awareness and letter knowledge). For the ID-DHH group, stimuli were repeated orally or in sign if necessary. For future studies in children with ID-DHH we recommend to directly start with oral stimulus presentation because the quality of sound from a laptop was too low for nearly all participants with ID-DHH.

The present study should be seen as a first attempt to gauge the reading level of children with ID-DHH and gain some directions for future studies in this field, because no research has been available so far. The study included only an ID and TD reference group of similar chronological age. This approach was chosen because education to children with ID-DHH in The Netherlands resembles the education for children with ID more than special education for children who are DHH. To further investigate the literacy of children with ID-DHH, it is necessary to include a reference group of children who are DHH as well. This will allow the inclusion of more tasks that are suitable for children who are DHH, which will offer more specific insight in how the severity of hearing loss affects literacy learning in children with ID-DHH.

The present results showed that the degree of intellectual disability or hearing loss was not decisive for any of the literacy levels. It is possible that more detailed information about hearing profile, age of implantation, preferred home language, sign and speech reading proficiency would have revealed some effects of their hearing loss (Johnson & Goswami, 2010; Knoors & Marschark, 2014). These factors are of great relevance for the literacy attainment of children who are DHH. However, it is uncertain to what extent they affect the literacy development of children with ID-

DHH, because their lower general cognitive level affects their general perception and rate of learning. More in-depth knowledge is also necessary about auditory discrimination, phonological and phonemic awareness, auditory and visual working memory, cognition (i.e. auditory and visual pattern recognition, reasoning) and word decoding skills in relation to reading in children with ID-DHH (Daza et al., 2015; Perfetti & Sandak, 2002). Finally, environmental factors such as home literacy and the type of reading instructions in schools may also contribute to language development in children with ID-DHH (Lederberg et al., 2013; Van der Schuit, Peeters, Segers, Van Balkom, & Verhoeven, 2009).

Conclusions and practical implications

The present study demonstrates the detrimental consequence of hearing loss combined with an intellectual disability for literacy attainment, each limiting different aspects. It also emphasizes the potential of some children with ID-DHH to attain basic levels of conventional literacy, as demonstrated by the small number of participants with ID-DHH who were able to perform at a basic level of conventional literacy. It is important to stay alert on the literacy potential of all children with ID-DHH, throughout childhood and provide access to language and literacy (for an educational rationale see Knoors & Vervloed, 2003).

The relationships we found between literacy skills and predictor skills are consistent with existing literature on children with TD, ID, and DHH. The level of IQ or hearing loss were not decisive for the level of literacy, suggesting that approaches to literacy should not focus on one disability over the other, and be based on both fields of expertise. The level of language skill (vocabulary in particular) was strongly related to reading comprehension, with decoding as a prerequisite. These results stress the importance of investing in language development for children with ID-DHH from an early age. Early intervention in language development may be greatly beneficial for literacy development in the long term.

Finally, it is important to keep in mind that each student has individual educational needs. The five participants with ID-DHH who were able to comprehend a simple text, were mainly distinguished by their decoding skills and vocabulary. However, on an individual level we found a unique combination of strengths and weaknesses that enabled each child to achieve literacy. This is an incentive to learn more about the optimal support that can be offered to children with ID-DHH, to expand their language and encourage their literacy. All children with ID-DHH will need a differentiated approach to provide support in a way that is tailored to their individual requirements.

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APPENDIX A

Additional Information on Participants with ID-DHH.

Table.

Continued

Participant	Age (months)	Gender	IQ	Hearing loss L (dB)	Hearing loss R (dB)	Hearing device	Language preference	Additional diagnoses
1	126	female	70	55	55	BAHA	Verbal	18Q (Grouchy) syndrome
2	166	male	81	90	90	None	SLN	CHARGE syndrome, Visual impairment (30%)
3	138	male	69	75	75	CI	SSD	Partial colourblindness, Spastic diplegia cerebral palsy
4	133	female	59	52	53	CI	SSD	Anxiety disorder, Attachment disorder
5	164	female	57	55	53	CI	Verbal	Hypersensitive to light, De Crouchy syndrome
6	114	male	64	70	70	1 BTE	Verbal	Chromomosome Xq21.1 deletion Dyspraxia
7	118	male	62	105	93	2 BTE	SSD	Motor problems, KISS syndrome
8	115	male	55	>80	>80	CI	SSD	Pneumococcal menigitis
9	119	female	65	95	105	CI	SSD	Anoxia at birth, Autism, Genetic mutation, Disturbed sensory processing
10	141	male	62	100	55	CI	Verbal	White matter anomaly
11	137	male	55	50	50	BAHA	Verbal	Autism, chromosomal abnormality 16q+ and 18q-
12	128	female	65	120	120	CI	SSD	Cytomegalovirus (CMV) infection
13	114	male	66	50	50	2 BTE	Verbal	Visual impairment, Zellweger syndrome
14	139	female	68	150	150	None	SLN	Autism disorder, Missing left vestibular, Epileptic disorder
15	123	female	60	80	100	CI	SSD	CHARGE syndrome, Visual impairment
16	102	female	72	unknown	85	None	SLN	Missing left eye
17	158	female	64	100	115	CI	SSD	Psychomotoric retardation , Microcephaly
18	110	male	70	52	53	2 BTE	Verbal	
19	166	female	54	50	40	2 BTE	SSD	
20	131	female	70	120	105	CI	SLN	Autism, CMV
21	150	male	63	65	>80	BTE+CI	SLN	Autism
22	102	female	50	80	40	2 BTE	SLN	Delayed motor deveopment, Mutiple operations to skull, Blocked tear ducts
23	138	male	65	80	120	1 BTE	Verbal	Aspects of PDD - NOS
24	161	female	69	100	110	CI	Verbal	Shah-Waardenburg syndrome, Mild visual disablity
25	130	male	70	105	105	2 BTE	Verbal	ADHD
26	138	male	68	83	83	BAHA	SSD	PDD-NOS Sensory processing issues
27	125	female	51	95	95	CI	SLN	Squint in left eye, lazy eye, CMV
28	116	female	57	70	70	2 BTE	Verbal	PDD -NOS
29	137	female	62	85	85	1 BTE	SSD	Noonan syndrome

Note. CI = Cochlear Implant, BTE = Behind The Ear hearing aid, BAHA = Bone Anchored Hearing Aid, SSD = Sign Supported Dutch, SLN = Sign Language of The Netherlands.

APPENDIX B

Nonparametric Tests and Effect Size of Within-Group Differences Between Participants with ID-DHH who Scored Below (Low) and Above (High) Chance Level on Phonological Awareness, Letter Knowledge, Decoding and Reading Comprehension.

	Phonological Awareness				**Letter Knowledge**				**Decoding**				**Reading Comprehension**			
	Mdn Low (*n* = 14)	**Mdn High (*n* = 11)**	***U***	***r***	**Mdn Low (*n* = 3)**	**Mdn High (*n* = 26)**	***U***	***r***	**Mdn Low (*n* = 16)**	**Mdn High (*n* = 13)**	***U***	***r***	**Mdn Low (*n* =10)**	**Mdn High (*n* = 5)**	***U***	***r***
Age	134.5	126	64	.14	137	130.5	33	.08	124.5	138	**60**[b]	.36	121	138	**9***	.51
IQ	67	62	**36***	.45	65	64	34.5	.06	64	65	100	.03	63	69	16	.29
Hearing loss left (dB)	86.5	80	58.5	.14	65	83	12	.23	75	85	78	.12	95	55	13	.34
Hearing loss right (dB)	102.5	72.5	43.5	.32	50	84	25	.16	62.5	85	71.5	.18	93	55	14.5	.29
Reading comprehension													6	14	**0****	.80
Decoding									10.1	30	**0****	.85	10.3	34.4	**3****	.70
Phonological awareness																
Rhyme	7	12	**12.5****	.71	8	8.5^{1}	11.5^{24}	.01	7^{12}	10	45.5	.36	10^{9}	9	22	.02
Synthesis	6	9	**26.5****	.56	7	7	25^{24}	.17	6.5^{14}	9	58	.31	7	13	10.5	.47
LetterKnowledge					14	31	**0****	.52	30	31	88.5	.13	31	33	18.5	.21
Vocabulary	62	78	53.5	.26	58	73.5	18.5	.27	60.5	78	73	.25	68.5	97	**6.5***	.59
Grammar	23^{13}	24	63.5	.10	17	23.5^{24}	11.5	.37	22^{15}	22^{12}	84	.06	24	27	15.5	.30
Listening comprehension	3^{13}	6	49.5	.26	4^{2}	6^{23}	19	.08	6^{12}	6	70.5	.08	5^{9}	10	8.5	.50
Rapid naming	38	36	68.5	.09	36	36.5	33.5	.07	34.5	37	75.5	.23	32	42	10	.47

Table.
Continued

Auditory perception																
Word repetition	0	5	**32.5***	.54	0	0	34.5	.07	0	0	85	.17	0.5	5	17.5	.25
Pseudoword repetition	0	0	48	.42	0	0	36.5	.04	0	0	76.5	.30	0	0	23.5	.05
Memory span																
DigitSpan	4^{10}	4	50.5	.07	2^{2}	4^{22}	9	.29	4^{11}	4^{13}	57	.18	4^{6}	4	11	.26
LetterSpan	3^{12}	3^{10}	47	.19	3	3^{23}	34	.01	2^{15}	4^{11}	57.5	.26	2.5	4	11	.46
Temporal processing	4.5	3	69	.09	2	4.5	38.5	.01	5.5	2	88.5	.13	5.5	12	21	.13
Nonverbal reasoning	20	19	64.5	.14	25	19.5	37.5	.02	19	25	78.5	.21	19.5	25	15	.32

Note. Deviating *n*'s are indicated behind *U*-scores in superscript.

$^{*}p_{exact} < .05$, $^{**}p_{exact} < .01$.
$^{a}p_{exact} = .051$. $^{b}p_{exact} = .53$.

6.

General Discussion

The aim of the present thesis was to explore the reading comprehension development of children with mild intellectual disabilities (ID) and of children with mild intellectual disabilities and additional hearing impairment (ID-DHH). The goal was to mark any specific characteristics in their pattern of reading development and to gain insight in their requirements for reading education. Correlational and longitudinal analyses were performed to investigate the relationship between reading comprehension and its main precursors at different levels. The reading systems framework (Perfetti, 1999; Perfetti & Stafura, 2014) was the overarching theoretical framework, with a focus on word decoding and listening comprehension, in line with the simple view of reading (Hoover & Gough, 1990). This chapter will summarize and interpret the main results, followed by a discussion on limitations and future directions. The chapter ends with a review of the practical implications for reading education of children with ID and ID-DHH.

Levels of Reading comprehension

The study presented in Chapter 2 examined the role of language comprehension, word decoding and vocabulary at explicit and implicit reading comprehension, when controlling for cognitive skill (nonverbal reasoning). Children with ID who had basic word decoding skills were compared to typically developing children who had the same level of word decoding.

The study showed that for lower-level reading comprehension (anaphora) the comprehension level was consistent with decoding level for both groups, while at higher-level comprehension (inference making) the ID group struggled to reach the same level as the typically developing control group notwithstanding their older age. The ID group also had lower levels of nonverbal reasoning and language comprehension. These findings indicate that cognitive difficulties especially affect the higher-level processes in reading comprehension, even when the general developmental delay of the reader is taken into account. Similar results have been found in individuals with ID of different ages and etiologies (Ezell & Goldstein, 1991; Facon, Facon-Bollengier, & Grubar, 2002; Jones, Long, & Finlay, 2006; Nash & Heath, 2011). Nonetheless, the strength of the relationship between nonverbal reasoning and higher-level reading comprehension was the same as for lower-level reading comprehension. We reasoned that additional cognitive abilities were related to higher-level reading comprehension that were not included in our study, such as working memory and other skills related to executive functioning (Schuchardt, Gebhardt, & Mäehler, 2010; Sesma, Mahone, Levine, Eason, & Cutting, 2009).

With regard to the general process of reading comprehension, the study is consistent with the simple view of reading (Hoover & Gough, 1990), since decoding and language comprehension skill were related to one or both reading comprehension measures (but stronger to lower level reading comprehension). Consistent with the convergent skills model (Vellutino, Tunmer, Jaccard, & Chen, 2007), decoding was an important predictor for reading comprehension in this group of early readers. Support for the lexical quality hypothesis (Perfetti, 2007) was less clear, since vocabulary was not identified as a main factor in reading comprehension. An explanation may be that they were still in the early stages of reading development and the integration of language-related reading skills has not yet occurred (Ouellette & Beers, 2010). However, we found a significant relationship between vocabulary and listening comprehension, suggesting an indirect effect on reading comprehension rather than a direct influence (in accordance with Language and Reading Research Consortium, 2015). Given this pattern of predictors, the simple view of reading (Hoover & Gough, 1990) was chosen as the basis for further research on reading comprehension in children with ID. The test battery was expanded with other cognitive predictors, as well as predictors for word decoding and listening comprehension

Individual Variation

Chapters 3 and 4 further explored the precursors of reading comprehension in a group of children with mild ID. The use of structural equation modeling in a one-year and a three-year analysis provided a more complete view on these precursors than was available so far. Both chapters yielded models that were highly similar to the typical predictors of reading: reading comprehension was related to word decoding and listening comprehension, consistent with the simple view of reading (Hoover & Gough, 1990). As in Chapter 2, word decoding was the main predictor for reading comprehension, consistent with the elementary level of reading that was applicable to the majority of the participants (Vellutino et al, 2007; Verhoeven & Van Leeuwe, 20008). The predictors that were found for word decoding (rapid naming, early literacy skills) and language comprehension (grammar, vocabulary) were also in line with earlier findings (Kintsch & Rawson, 2005; Melby-Lervåg, Lyster, & Hulme, 2012). Again as in Chapter 2, vocabulary in Chapter 3 and 4 was only related to language comprehension and not to word decoding or reading comprehension, consistent with the pattern in early readers (Ouellette & Beers, 2010; Verhoeven & Van Leeuwe, 2008). In early readers, reading comprehension is not (yet) fully integrated into the language comprehension network, but is strongly dependent on word decoding skills (Landi, Frost, Menc, Sandak, & Pugh, 2013; Vellutino et al., 2007). In addition, word decoding in transparent orthographies is less dependent on oral vocabulary than in complex orthographies (Seymour, Aro, & Erskine, 2003). Taken together, the present results represent a structure of predictors that is consistent with the typical reading comprehension pattern of children in the early grades.

At the same time, several additional direct relationships were found in the children with ID where only indirect relationships would be expected in a typical situation. Remarkable were the additional relationships between reading comprehension and early literacy skills, and between reading comprehension and cognitive predictors (temporal processing and nonverbal reasoning). A direct connection between early literacy skills and reading comprehension, on top of word decoding, indicates that phonological deficits are a main reason for reading problems in children with mild ID. The little available literature suggests that scores on phonological awareness may reflect metacognitive skills such as working memory and metacognitive awareness (Engen & Høien, 2002). Although the present research was not set up to look further into the nature of this relationship, an additional multiple regression analysis confirmed that working memory and nonverbal reasoning accounted for 54% of the variance in the composite score of early literacy skills in Chapter 3.

The regression analysis in Chapter 4 revealed several additional cognitive precursors underlying the main linguistic skills in reading comprehension; rapid naming, pseudo-word repetition, working memory and nonverbal reasoning related to reading comprehension, word decoding and language comprehension. Most of these cognitive predictors were no longer significant when linguistic predictors were added to the model, which implies an indirect effect of these cognitive skills on reading comprehension rather than a direct effect. These observations are consistent with the claims of the reading systems framework (Perfetti & Stafura, 2014). However, both in Wave 1 as when taking a longitudinal perspective we found additional direct relationships between cognitive skills and reading comprehension, in addition to linguistic predictors. For typically developing children, several longitudinal studies have reported a direct effect of verbal working memory and analogical reasoning on reading comprehension when the autoregressor was taken into account (Seigneuric & Ehrlich, 2005; Stevenson, Bergwerff, Heiser, & Resing, 2014) and even when controlling for word decoding and language comprehension (Fuchs, Compton, Fuchs, Bryant, Hamlett, & Lambert, 2012). Direct relationships between temporal processing and reading comprehension have also been found for auditory and visual-auditory stimuli, next to a mediating effect of phonological awareness (Malenfant, Grondin, Boivin, Forget-Dubois, Robaey, & Dionne, 2012). These findings underline that cognitive processing abilities are directly applied during the reading comprehension process. They also suggest that problems in reading comprehension are partly caused by limitations in the processing and interpretation of sensory stimuli.

The impact of hearing impairment

In Chapter 5, the research in the previous chapters was applied to children with ID who are also deaf or hard of hearing (ID-DHH). They scored significantly lower than the ID reference group on nearly all tasks, cognitive as well as linguistic, although they had similar level of IQ and nonverbal reasoning. There was wide variability in individual reading profiles, but several observations could be made at group level. Generally speaking, better readers were distinguished by age, word recognition and vocabulary. In addition, a relationship was found between reading comprehension and listening comprehension, nonverbal reasoning, rapid naming and short-term memory. The relationships that were found between decoding, reading comprehension and predictor skills were consistent with existing literature on children with typical development and ID with regard to the simple view of reading (Hoover & Gough, 1990), the lexical quality hypothesis (Perfetti, 2007) and the convergent skills model (Vellutino et al., 2007). The relationship of reading comprehension with reasoning and memory measures was consistent with findings in Chapters 2, 3, and 4 on children with mild ID.

Several observations at group level were clearly consistent with the reading development of children who are DHH. First, although the children's level of letter knowledge and decoding was reasonable, phonological awareness was weak and not visibly related to reading scores in this group. This implies that word reading at this early stage was mainly achieved through direct orthographic recognition, consistent with the reading development of children who are DHH (Kyle & Harris, 2010). Second, where vocabulary is not seen as a primary predictor of reading comprehension in typical early readers and in early readers with ID, in Chapter 5 it was a notable indicator of reading comprehension level for children who were ID-DHH. This agrees with the notion that children who are DHH use word meanings to connect orthographic word representations to sign language (Hermans, Knoors, Ormel, & Verhoeven, 2007). Third, while basic decoding was above chance level for 13 out of 29 participants, elementary reading comprehension was observed in only 5 participants. This is in line with the reading profile of children who are DHH, who have a limited depth of lexical knowledge and may not be familiar with the grammar and structure of written language in comparison to sign language (Coppens, Tellings, Van der Veld, Schreuder, & Verhoeven, 2012; Lederberg, Schick, & Spencer, 2013).

The results point to a combined influence of both impairments: characteristics of reading development in children with a hearing impairment were observed in combination with the consequences of an intellectual disability. Importantly, there was a small group of participants who had gained a basic level of reading comprehension. Their number might be increased if reading education is tailored to the specific needs of children with ID-DHH. Based on the present study we can say that word recognition is necessary, but not sufficient for reading comprehension in children with ID-DHH. A general understanding of spoken language and vocabulary is essential for reading comprehension in children with ID-DHH as well.

Reading comprehension in children with ID – revisited

With regard to the theoretical framework, the present study yielded results that are largely in line with the reading systems framework (Perfetti, 1999; Perfetti & Stafura, 2014). The study showed a typical pattern of predictors for reading comprehension in children with mild ID, with small additions to this framework. The simple view of reading (Hoover & Gough, 1990) was an adequate model for their reading comprehension process. For children with ID-DHH, support for the simple view of reading was found in individual patterns, along with within-group differences between good and weak reading comprehenders. Support for the lexical quality hypothesis (Perfetti, 2007; Perfetti & Hart, 2002) was limited in children with mild ID. There was only a link between vocabulary and listening comprehension and no direct relationship between vocabulary and decoding or reading comprehension. This may be due to their reading level, since the pattern was consistent with early readers. Further research would be necessary to investigate this relationship for children with ID with higher levels of word decoding. Reversely, in children with ID-DHH vocabulary was directly related to reading comprehension, but not with listening comprehension or decoding. However, the sample of participants was highly diverse in this study. A larger sample might have given stronger support for other relationships within the lexical quality hypothesis.

Regarding levels of reading comprehension, it was found in Chapter 2 that lower-level reading comprehension was more directly related to linguistic skills than higher-level reading comprehension. This pattern is consistent with the claim of the reading systems framework (Perfetti & Stafura, 2014) that lower-level linguistic skills are more strongly related to constructing a semantic representation of the text while higher-order cognitive skills are necessary for inference drawing to extract implicit information.

As expected, we found that a mild ID primarily affects the linguistic and general knowledge base, resulting in a lower general reading level. The stable pattern of linguistic predictors strengthens the proposition that weaknesses in reading comprehension are caused by cognitive factors underlying linguistic skills. The cognitive limitations in children with mild ID especially restricted performance in reading processes that require high cognitive load. Next to these indirect effects, the models in Chapter 2, 3 and 4 also demonstrate a small direct effect of cognitive skill on reading comprehension. For children with ID-DHH, these relationships were also found, with an additional deficiency in the orthographic and linguistic knowledge systems which further lowered their level of decoding and reading comprehension.

Limitations and future directions

There are some limitations of the present research that are important to address. First, the choice of control groups was based on chronological age rather than cognitive age or reading age level. As a result, the reading comprehension level was highly variable from the start of the study. However, chronological age was the only factor that would remain stable in a longitudinal design. Matching on reading level or cognitive age would have raised practical issues due to the large distribution of reading levels within the ID group(s), and the difference in learning rate which would still have resulted in a wide gap between groups from the second year onwards.

Second, the wide variability of reading skill and etiologies within the ID group was included deliberately, to obtain a realistic indication of the children that are in the classrooms within special education. The small amount of reading research that has been done so far, habitually used a select group with specific impairments, such as Down syndrome. However, specific syndromes are often paired with specific reading profiles. The present study focused on common characteristics in a general group of children with mild ID, to provide a basis from where to adapt the reading education of each student to their specific needs.

Third, the present study focused on literacy-related skills that were present in the participants and how these skills contributed to reading comprehension. However, many environmental factors are also influence the development of literacy and language comprehension, such as home literacy environment (Heath et al., 2014; Sénéchal & LeFevre, 2002; Van der Schuit, Peeters, Segers, Van Balkom, & Verhoeven, 2009) and the method for reading education in schools (Hill, 2016). More knowledge is needed regarding the effects of these factors separately, and about their combined effects on the development of reading comprehension in children with ID.

It is also valuable to increase knowledge on the effects of co-occurring disabilities or conditions on the reading development of children with ID. The present study made a first effort to investigate hearing impairment in combination with mild ID, but there are many other conditions that are common among the ID population that are known to complicate the development of language or literacy. It may be worthwhile to look further into the role of additional diagnoses in children with ID, in order to provide a more specific tailored educational program for subgroups of children with ID.

In addition, future studies could dig deeper in the effect of specific cognitive skills on reading, such as attention, executive functioning, or different forms of working memory. The present research demonstrates the importance of cognitive skills for reading comprehension in children with ID, both indirectly through language skills and directly in addition to language skills. Previous research has demonstrated that different aspects of temporal processing, working memory or executive functioning have different degrees of influence on reading comprehension (Malenfant et al., 2012) and the relative strength of cognitive skills changes over time (Davidson, Amso, Anderson, & Diamond, 2006).

Finally, it may be valuable to look into different methods to measure reading comprehension in children with ID. For typically developing children, it has been established that different types of tests tap into different sets of skills (Keenan, Betjemann, & Olson, 2008). Multiple-choice tests (as were used in the present study) intrinsically require working memory and language skills in order to perform the task of choosing between options. There may be other alternative comprehension tests with less cognitive load on the task itself, that may be better suited for children with ID.

Practical implications

The present research has shown that the cognitive deficits in children with mild ID influence language and reading comprehension at multiple levels. Based on the current results, several implications regarding reading education in children with ID can be mentioned.

With regard to the process of reading comprehension, an ID particularly affects word decoding and the higher-level comprehension processes. The attainment of

word decoding skills can be a laborious process for children with ID. However, several studies have shown that children with mild or moderate ID are able to learn phonics when they receive systematic, explicit instructions (Joseph & Seery, 2004; Hill, 2016). For example, Ainsworth, Evmenova, Behrmann, & Jerome (2016) showed that children with moderate and severe ID and complex communication needs made progress when given phonics instructions that were direct, systematic, highly structured, and offered in small groups. But even prior to formal literacy instruction there is also a wide variety of literacy experiences that can help a child to create a concept of written text and to learn that written language is a part of daily life. Informal encounters with text can be encouraged by providing access to reading materials, labeling objects and early literacy activities such as telling stories or singing songs, to stimulate phonological awareness. This can be extended further by using written text on conversation posters, making mind maps with words and concepts, and playing language games.

In reading comprehension, higher-level comprehension was the most problematic. Still, good inference making requires a good understanding of the content of the text. It is therefore important that children are familiarized with the process of constructing a text representation from the start of literacy education, and that their confidence in text reading is built. It is recommended to explicitly teach and model the steps for constructing a text representation from an early point in literacy education. When word decoding is still weak, students may benefit from simplification of a text to compensate for the effort that is needed for the decoding process. Persons with ID achieve a higher level of overall text comprehension when provided with short texts that require little inference making (Fajardo, Ávila, Ferrer, Tavares, Gómez, & Hernández, 2014). A second method to increase reading comprehension is the use of illustrations that help to construct a text representation and reduce cognitive load. Although symbol reading or the use of key word symbols does not necessarily help the comprehension of simple texts (Jones, Long, & Finlay, 2007; Poncelas & Murphy, 2007), it can be helpful to add illustrations that capture the overall meaning of a sentence or paragraph (Shurr & Taber-Doughty, 2012). When word decoding is more advanced, interventions can focus on teaching higher-level reading comprehension strategies (Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2014; Browder, Hudson, & Wood, 2013). For example, a group of 40 adolescents with mild ID showed improvement in reading comprehension and a more active involvement with the text after practicing four reading comprehension strategies: prediction, clarification, question generation and summarization, in a reciprocal teaching setting (Lundberg & Reichenberg, 2011).

The importance of vocabulary and language comprehension for reading comprehension was particularly visible in children with ID-DHH in the present study. Their reading pattern showed several similarities to children who are DHH, in addition to the effects of their cognitive limitations. Recommendations from both areas of expertise must be carefully considered when designing a literacy curriculum for children with ID-DHH. Specifically, it is recommended to strengthen both the breadth and depth of vocabulary by actively building on the semantic network. This can be done by repeated explicit and implicit encounters with new words, defining and explaining word meanings, and encouraging deep and active processing of word meanings in a range of contexts (Luckner & Cooke, 2010). For children with ID-DHH it is especially important to learn concepts in meaningful, experience-based learning environments that relate to their personal interests and understanding (Van der Schuit, Segers, Van Balkom, Stoep, & Verhoeven, 2010). Language skills can be stimulated further by engagement in conversations and literacy activities. For example, shared picture book reading is beneficial to many aspects of vocabulary and language development of children who are DHH (Luckner, Bruce, & Ferrell, 2016) and children with ID (Davie & Kemp, 2002).

In preference to providing systematic instructions for all literacy skills separately, it is beneficial to use an integrative approach. Several studies have reported successes of comprehensive reading instruction that integrate all elements of reading. For example, Allor et al. (2014) provided systematic and explicit comprehensive reading instruction to children of different levels of ID (IQ between 40 and 80) from Grade 1 through 4. Daily instructions were given in groups of 1-4 students. The lessons included reading activities at word level, fluency, and comprehension. Over all IQ levels, children who followed this curriculum made greater progress on measures of phonological awareness, decoding fluency, and reading comprehension than children in a control group that received the usual instruction. An example of an intervention that had a clear multi-modal approach is described by Coyne, Pisha, Dalton, Zeph, & Smith (2012). The method focused on reading for meaning and provided direct instruction in decoding and comprehension skills in a multi-modal digital environment.

In addition to linguistic precursors, the present research also shows the importance of cognitive skills in reading comprehension. Cognitive skills are necessary for the perception and processing of speech and text, but are also directly involved in top-down reasoning and constructing a meaningful representation of the text. Reading education may therefore also benefit from activities that specifically stimulate the improvement of cognitive precursor skills such as pattern recognition, visual matching, attention, reasoning and the perception and production of rhythm.

As a final point, in the present thesis it was observed that decoding and reading comprehension skill in children with ID and ID-DHH improved over time, albeit slowly. More studies have shown that success in decoding may start later in education and continues in the higher grades (e.g., Lemons et al., 2013). In the study by Allor et al. (2014) children with mild ID on average needed three academic years to gain the average reading fluency of first grade. Regrettably, it is not unusual that schools in The Netherlands resort to teaching only sight-word reading when a set period of instruction does not lead to noticeable reading development. The observed reading gains in older students with ID are an encouragement to keep providing phonics based reading instruction (possibly alongside other methods) if a student does not learn to decode in the first years of reading education. In addition, the slow learning curve can be taken as a motivation to start early with literacy experiences in a playful manner such as songs, shared book reading and telling stories. These activities may prime the child's sensitivity for print and story structure and may help to develop literacy skills at a later moment.

In conclusion, the present research shows that a majority of children with mild ID and children with ID-DHH are able to achieve elementary reading comprehension. The development of decoding and reading comprehension is hindered by problems in information processing and cognition, but overall their reading profile is similar to typically developing readers. Reading education should be designed accordingly, by providing a learning environment that stimulates the development of cognitive and linguistic precursor skills for reading comprehension and provides support in the processing and interpretation of spoken and written language. A highly structured, integrative and comprehensive approach can help children with ID to reach their full potential in reading comprehension.

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Nederlandse samenvatting

Dankwoord

Curriculum Vitae

Nederlandse samenvatting

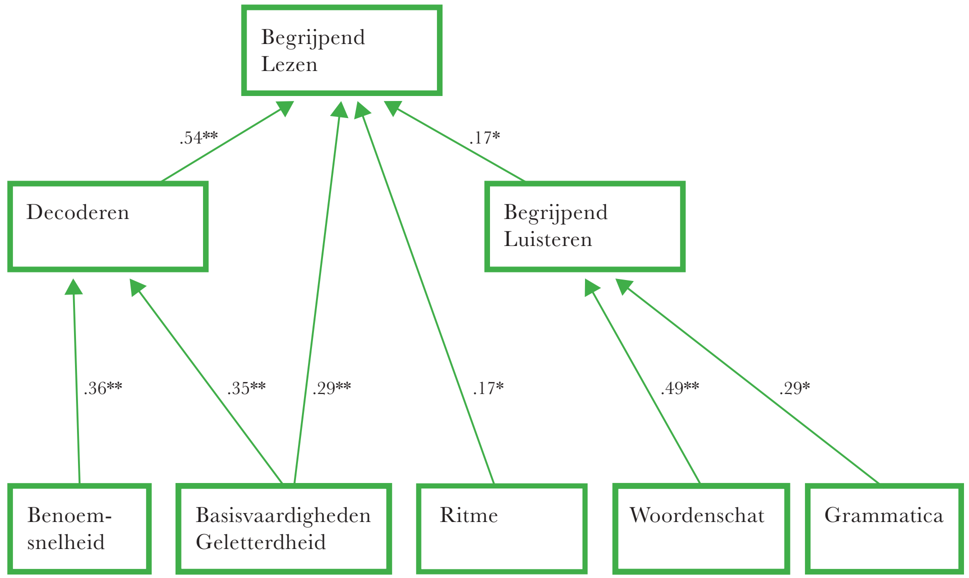
Kinderen met een verstandelijke beperking (VB) hebben vanwege hun cognitieve problemen vaak moeite met het leren van nieuwe, abstracte vaardigheden. Voor het lezen en begrijpen van een tekst zijn veel van deze cognitieve vaardigheden nodig. Bovendien zijn er veel taal-en begripsprocessen bij betrokken. Zo moet de lezer geschreven woorden vertalen naar hun gesproken variant, en vervolgens de betekenis van de tekst begrijpen (Hoover & Gough, 1990). Hiervoor is ook een gedegen woordenschat van belang: zowel het kennen van veel woorden, als het goed kennen van woorden draagt bij aan tekstbegrip (Perfetti, 2007). Ten slotte moet de lezer niet alleen de letterlijke betekenis van de tekst construeren, maar ook de diepere, impliciete betekenis begrijpen (Kintsch & Van Dijk, 1978). Om tot een goed tekstsbegrip te komen zijn veel basiskennis en -vaardigheden nodig, zoals letterkennis en fonologisch (klank)bewustzijn, maar ook werkgeheugen, redeneervermogen en verwerkingssnelheid. Bij kinderen met een VB zijn deze vaardigheden echter vaak in mindere mate aanwezig, waardoor zij ook problemen ondervinden bij het ontwikkelen van leesvaardigheid en bij het begrijpend lezen. Veel kinderen met een VB hebben bovendien een zintuigelijke beperking. Kinderen met een VB zijn bijvoorbeeld zes keer vaker dan gemiddeld doof of slechthorend (D/SH-VB). Hun toegang tot gesproken taal is vaak beperkt, wat een extra barrière kan zijn voor het leren lezen en begrijpen van tekst. Het is dus extra belangrijk dat deze groepen goed ondersteund worden in hun leesleerontwikkeling. Er is echter weinig wetenschappelijk onderzoek beschikbaar naar de voorspellers voor begrijpend lezen bij kinderen met een VB en er is nog geen enkele studie gedaan naar dit onderwerp bij kinderen met D/SH-VB. In het huidige onderzoek is gekeken naar de onderlinge samenhang tussen vaardigheden die nodig zijn voor begrijpend lezen in kinderen met een VB en kinderen met D/SH-VB. Het doel was om te bepalen in hoeverre het leespatroon van deze doelgroepen afwijkt van het patroon bij zich normaal ontwikkelende kinderen. De resultaten kunnen worden gebruikt voor het optimaliseren van de leesleertrajecten van kinderen met een VB of D/SH-VB.

Lezen bij kinderen met een verstandelijke beperking

Hoofdstuk 2 beschrijft een onderzoek naar een selectie van de belangrijkste voorspellers voor begrijpend lezen bij kinderen met een normale ontwikkeling: decoderen, begrijpend lezen, woordenschat en nonverbaal redeneren. De studie liet zien dat kinderen met een verstandelijke beperking (VB) die een basaal leesniveau hadden (groep 3-niveau) dezelfde mate van expliciet tekstbegrip hadden als zich normaal ontwikkelende leerlingen met dezelfde decodeervaardigheid. Zij waren

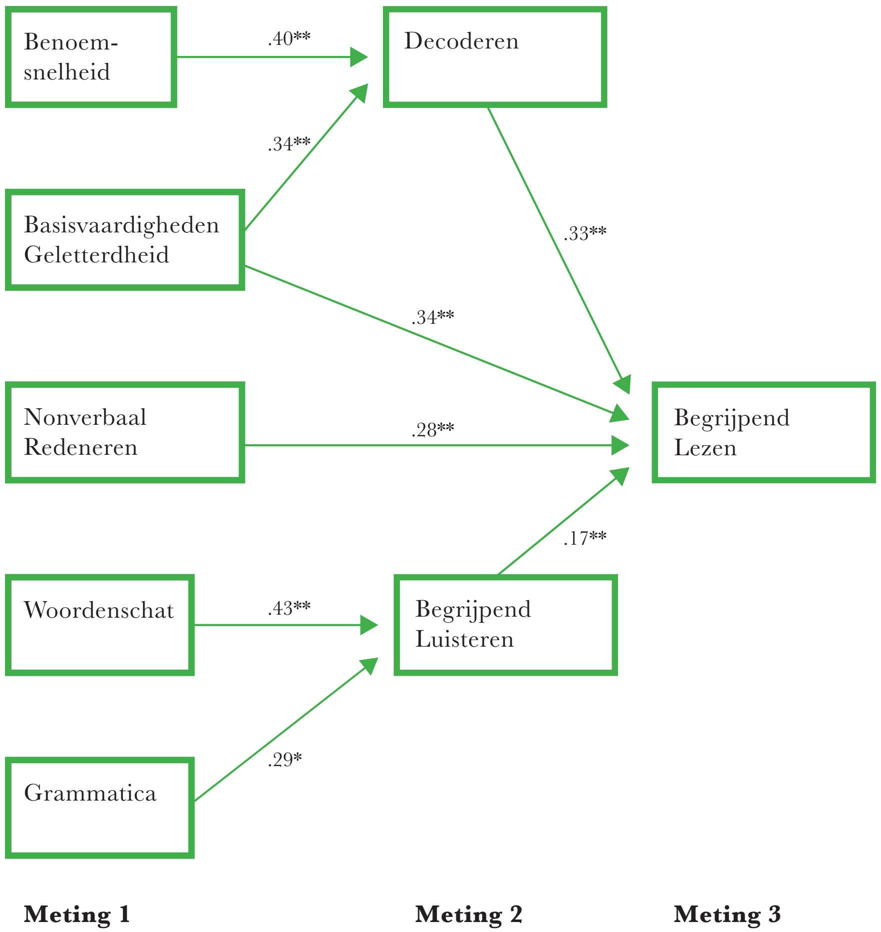
echter zwakker op het gebied van nonverbaal redeneren en impliciet tekstbegrip. Het expliciet tekstbegrip was gerelateerd aan zowel decoderen als begrijpend luisteren, net zoals bij normaal ontwikkelende kinderen. Dit patroon is consistent met één van de belangrijkste theorieën over begrijpend lezen, de Simple View of Reading (Hoover & Gough, 1990). Voor impliciet tekstbegrip was decoderen een belangrijke predictor voor de score op de taak, maar begrijpend luisteren niet. Hier leken nog andere cognitieve vaardigheden een rol te spelen, die in dit onderzoek niet meegenomen waren.

In hoofdstuk 3 en 4 zijn meer cognitieve variabelen meegenomen, zoals werkgeheugen en nonverbaal redeneren Hiervoor zijn 81 leerlingen met een verstandelijke beperking (11,5 – 12,5 jaar oud, IQ 50 – 80, gemiddeld IQ 60.4) en 86 zich normaal ontwikkelende leerlingen (10,5 – 11,5 jaar oud) getest. Het doel was om te achterhalen wat het patroon van leesontwikkeling is bij kinderen met een VB. Met behulp van Structural Equation Modeling (SEM) is op meetmoment 1 en 3 een model ontwikkeld om de onderlinge relaties te beschrijven tussen voorspellers voor begrijpend lezen. Het model op meetmoment 1 is weergegeven in Figuur 1 en het longitudinale model na meetmoment 3 is weergegeven in Figuur 2.



Figuur 1.
Model met cognitieve en linguïstische predictoren voor begrijpend lezen bij kinderen met een verstandelijke beperking op meetmoment 1.

App.



Figuur 2.
Longitudinaal model met de cognitieve en linguïstische predictoren op meetmoment 1, decoderen en begrijpend luisteren op meetmoment 2, en begrijpend lezen op meetmoment 3.

De kinderen lieten een grote variatie aan leesvaardigheid zien, maar een meerderheid kwam wel tot een basisniveau van geletterdheid. De modellen op moment 1 en 3 zijn in veel opzichten gelijk. Bovendien zijn ze consistent met het beeld dat zich normaal ontwikkelende leerlingen laten zien in de lagere klassen van het basisonderwijs. Begrijpend lezen wordt in sterke mate voorspeld door decoderen (voorspeld door benoemsnelheid en de basisvaardigheden voor geletterdheid: letterkennis en fonologisch bewustzijn) en in mindere mate door begrijpend luisteren (wat weer voorspeld wordt door woordenschat en grammaticaal begrip). Opvallend is verder

de directe relatie tussen de basisvaardigheden voor geletterdheid en begrijpend lezen, *naast* de relatie die via decoderen loopt. Het is mogelijk dat deze relatie een representatie is van enkele cognitieve vaardigheden zoals werkgeheugen en een algemeen begrip van taal. Ook was er in beide modellen een directe relatie tussen begrijpend lezen en een cognitieve vaardigheid: het reproduceren van ritmes op meetmoment 1 en nonverbaal redeneren op meetmoment 3. Dit kan betekenen dat problemen met begrijpend lezen deels veroorzaakt worden door problemen met het verwerken of interpreteren van zintuiglijke waarnemingen, zowel visueel als auditief.

In het kort laten deze bevindingen zien dat problemen bij tekstbegrip bij kinderen met een VB deels veroorzaakt worden door beperkingen in de ontwikkeling van taal-gerelateerde vaardigheden: woordenschat, grammatica en de basisvoorwaarden voor geletterdheid. Echter, er is ook een direct effect van de cognitieve beperking op het begrijpend lezen, met name ritmewaarneming en nonverbaal redeneren.

Studie naar lezen bij kinderen met D/SH-VB

Hoofdstuk 5 richtte zich op het in kaart brengen van het leesniveau van leerlingen die naast hun VB ook Doof of Slechthorend zijn (D/SH-VB). Deze studie richtte zich op het onderscheiden van mogelijke kind-kenmerken voor de ontwikkeling van geletterdheid, met het oog op de inrichting van het leesonderwijs voor deze doelgroep. Er namen 29 leerlingen deel met D/SH-VB in de leeftijd van 8,5 – 13,5 jaar (IQ 50 – 81, gemiddeld 63.6). Hun gehoorverlies was > 35 dB in het beste oor. De bevindingen in deze studie zijn gerelateerd aan zowel de resultaten van het longitudinale onderzoek bij kinderen met een VB, als aan de bestaande kennis over het leesleertraject bij kinderen die D/SH zijn.

De resultaten benadrukken het grote effect dat D/SH-VB kan hebben op de taalontwikkeling en het leren lezen. De D/SH-VB groep scoorde lager dan de kinderen met alleen een VB op nagenoeg alle taken, behalve nonverbaal redeneren, hoewel beide groepen nagenoeg hetzelfde IQ hadden. Ondanks dat zij woorden konden herkennen, scoorden de meeste kinderen met D/SH-VB onder kansniveau op test-items voor begrijpend lezen. De kinderen die het beste scoorden op begrijpend lezen waren over het algemeen ouder dan de kinderen die lager scoorden op deze taak. Dit betekent echter niet dat pas op oudere leeftijd begonnen zou moeten worden met leesonderwijs; veeleer lijkt het erop dat deze leerlingen een lange aanloop nodig hebben. De tweede belangrijke indicator voor begrijpend lezen bleek woordherkenning. Het leek er echter op dat een deel van de kinderen deze taak uitvoerde op basis van directe woordherkenning en niet door de woorden spellend te lezen (decoderen). Alleen bij de kinderen die de vaardigheid van decoderen wel onder de knie hadden, was hun leesvaardigheid flexibel genoeg om ingezet te worden bij het begrijpen van een onbekende tekst. Tenslotte was de algemene taalontwikkeling een belangrijke factor in het leesniveau van kinderen met D/SH-VB. Woordenschat wordt als belangrijke voorwaarde gezien voor de leesontwikkeling van zich normaal ontwikkelende en D/SH kinderen, begrijpend luisteren wordt ook gevonden als voorspeller voor tekstbegrip bij kinderen met een VB. In het kort wijzen deze resultaten erop dat –in het kader van leesonderwijs- niet één van beide beperkingen als dominant moet worden beschouwd, maar dat het belangrijk is om vanuit beide perspectieven te werken.

Praktische implicaties

Het huidige onderzoek heeft laten zien dat een VB het leesleertraject van kinderen op verschilldende manieren beïnvloedt. Ten eerste hebben veel kinderen met een VB moeite met het leren lezen op zich. Ook op het gebied van begrijpend lezen hadden zij in dit onderzoek een achterstand, met name bij het begrijpen van impliciete informatie.

Wat betreft leren decoderen is bekend dat kinderen met een VB baat hebben bij systematische, expliciete instructie die wordt aangeboden in kleine groepjes (Ainsworth, Evmenova, Behrman, & Jerome, 2016; Hill, 2016). Maar zelfs voorafgaand aan het formele leesonderwijs kunnen zij al geholpen worden om hun gevoeligheid voor geschreven tekst te ontwikkelen. Door hen actief te betrekken bij geletterdheids-activiteiten (zoals voorlezen, liedjes zingen en het actief uitbreiden van de woordenschat), maar ook door indirecte ervaringen de beschikbaarheid van boeken en observatie van anderen die geletterdheid gebruiken in het dagelijks leven.

Ook voor het begrijpend lezen is goede begeleiding nodig voor kinderen met een VB. Het is belangrijk dat kinderen met een VB strategieën aangereikt krijgen om tot een goed tekstbegrip te komen. Net zoals voor decoderen is op dit gebied een systematische, expliciete instructie aan te raden (Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2014). Bij voorkeur wordt er gebruik gemaakt van een geïntegreerde aanpak, waarbij decoderen, taalvaardigheid en begrijpend lezen gecombineerd worden aangeboden. Op deze manier leren kinderen verbanden leggen tussen de verschillende vormen van taal (geschreven en gesproken) en krijgt het lezen betekenis.

Voor kinderen die nog niet zo goed lezen kan het tekstbegrip op een aantal manieren ondersteund worden. Ten eerste kunnen illustraties helpen bij het begrijpen van

moeilijke woorden en het leggen van verbanden binnen de tekst. Voor begrijpend lezen hebben kinderen met een VB bijvoorbeeld baat bij illustraties die de betekenis van een tekstgedeelte verbeelden (Shurr & Taber-Doughty, 2012). Ook is het van belang om de teksten voldoende te vereenvoudigen zodat zij genoeg cognitieve capaciteit kunnen inzetten voor het overzien van de tekst. Een korte tekst met weinig impliciete informatie kan helpen bij het leren begrijpend lezen. Zodra het leesniveau hoger wordt, kan ook de moeilijkheid van de teksten in kleine stappen worden aangepast en kunnen complexere leesstrategieën worden aangeleerd.

Voor kinderen met D/SH-VB is bovendien aan te bevelen om allereerst in te zetten op ontwikkeling van de algemene taalvaardigheid en kennis van de gesproken taal, naast de actieve opbouw van andere communicatievormen zoals gebaren of picto's. Daarbij hoort ook het aanbieden van letters en de instructie in fonologische vaardigheden vanaf jonge leeftijd. Een vergroting van de woordenschat (zowel verbreding als verdieping) en taalkennis vormt de basis voor het leren begrijpen van de geschreven taal. Taalvaardigheid kan gestimuleerd worden door de aanwezigheid van een taalrijke omgeving en extra stimulans door voorlezen, en het voeren van gesprekjes. Voor uitbreiding van de woordenschat is veel herhaling nodig, in combinatie met gerichte ondersteuning bij het begrijpen van nieuwe woorden en het inbedden van nieuwe begrippen in het semantisch netwerk. Zorg bij voorkeur voor een ervarings- of belevingsgerichte context voor het leren kennen van nieuwe begrippen (Van der Schuit, Segers, Van Balkom, Stoep, & Verhoeven, 2010).

Conclusie

Lezen kan kinderen een nieuwe mogelijkheid bieden om de wereld te ontdekken en hun ontwikkeling te stimuleren. Voor kinderen met een VB en D/SH-VB verloopt het leren lezen moeizamer dan bij de meeste andere kinderen. Het huidige onderzoek laat echter ook hun mogelijkheden zien, al duurt het soms langer voordat de geletterdheid op gang komt bij kinderen met een VB. Het is daarom van belang om kansen te blijven bieden voor het ontwikkelen van klankbewustzijn, decoderen en begrijpend lezen, eventueel naast alternatieve methoden. Gerichte, gestructureerde instructies, aansluitend bij hun interesses en leefwereld, kan kinderen met een VB helpen om hun mogelijkheden tot geletterdheid zo goed mogelijk te benutten.

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Curriculum Vitae

Evelien van Wingerden – Fontein (1986) behaalde haar VWO-diploma aan het Herbert Vissers College in Nieuw-Vennep met het profiel Natuur & Gezondheid en een profielwerkstuk over visuele waarneming. Hierna volgden een bachelor Psychologie en research master Cognitive Neuropsychology aan de Vrije Universiteit in Amsterdam, waarbij zij de nadruk legde op cognitive science. Van 2011 tot 2016 deed Evelien onder begeleiding van Ludo Verhoeven, Hans van Balkom en Eliane Segers promotie-onderzoek naar de ontwikkeling van begrijpend lezen bij kinderen met een verstandelijke beperking en kinderen die naast een verstandelijke beperking ook doof of slechthorend zijn. Daarnaast gaf zij werkgroepen academische vaardigheden en begeleidde zij verschillende scriptiestudenten van zowel de bachelor als de master Pedagogische Wetenschappen. Bij één van deze scriptie-onderzoeken werkte zij samen met Nina Wolters-Leermakers en Rita Gerkema-Nijhof (Kentalis) aan een effect-onderzoek naar de voorleesmethode Zintuigenverhalen voor kinderen met een communicatief meervoudige beperking. Momenteel werkt Evelien binnen de projectgroep 'Sociale relaties en ICT' van Paula Sterkenburg (VU) aan de ontwikkeling en effectstudie van de HiSense APP MVB+LVB, bedoeld voor ouders en begeleiders van personen met een matige of lichte verstandelijke beperking.