Signs for Developing Reading

Sign Language and Reading Development in Deaf and Hard-of-Hearing Children

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Abstract

Reading development is supported by strong language skills, not least in deaf and hard-of-hearing (DHH) children. Regional Special Needs Schools (RSNS) in Sweden have a bilingual curriculum, that is, DHH pupils at these schools learn Swedish Sign Language and written and/or spoken Swedish. The work in the present thesis investigates reading development in DHH children who attend these schools and who are learning to read. The primary aim of the present work was to investigate whether the reading skills of DHH signing children can be improved via computerized sign language based literacy training aimed at strengthening the connections between sign language and written language. Another aim was to investigate concurrent and longitudinal associations between skills in reading, sign language, and cognition in this population. The results indicate that sign language skills support developing reading skills in RSNS pupils (Paper I and Paper IV). In particular, sign language based literacy training appears to support development of word reading (Paper IV). In addition, awareness of the sub-lexical structure of sign language seems to assist word reading (Paper I). Interestingly, the ability to imitate manual gestures was related to reading skills. More specifically, precision of imitation of unfamiliar signs was associated with development of word reading (Paper IV) and precision of imitation of familiar signs, i.e., vocabulary, seemed to be associated with developing reading comprehension (Paper IV). Results also suggest that working memory and Theory of Mind (ToM) are related to reading comprehension in RSNS pupils (Paper III). In addition to these findings relating to reading development, results also suggest that sign language experience enhance the establishment of representations of manual gestures (Paper II), and that progression in ToM seem to be typical, although delayed, in RSNS pupils who are learning to read (Paper III). Taken together, the associations revealed between sign language skills and reading development support the notion that sign language skills may provide a foundation for promotion of reading skills in DHH signing children. Thus, interventions that support development of sign-based representations and their manipulation and use in written language processing, may improve reading skills in this population. To account for the present findings, a model of written language processing is proposed. Working memory has a central role in integrating environmental stimuli and language-mediated representations, and thereby provides a platform for cross-modal language processing and multimodal language development.

Sammanfattning

En god språklig förmåga bidrar till god läsutveckling, inte minst hos döva och hörselskadade barn. Specialpedagogiska skolmyndighetens specialskolor för döva och hörselskadade (D/H) barn har en tvåspråkig läroplan som innebär att elever på dessa skolor lär sig både svenskt teckenspråk och skriven och/eller talad svenska. De fyra arbeten som ingår i avhandlingen undersöker läsutveckling hos D/H elever på dessa skolor. Det huvudsakliga syftet var att undersöka om läsförmågan hos D/H elever som använder sig av teckenspråk förbättras via datoriserad teckenspråksbaserad lästräning som syftar till att stärka sambanden mellan teckenspråk och det skrivna språket. Ett annat syfte var att undersöka samtida och longitudinella samband mellan läsförmåga, teckenspråk, och kognition. Resultaten visar att kunskaper i teckenspråk kan stödja läsutveckling hos dessa elever (Artikel I och Artikel IV). Teckenspråksbaserad lästräning tycks bidra till ordläsningsutveckling (Artikel IV) och medvetenhet om teckenspråkets sublexikala struktur stöttar ordläsning (Artikel I). Förmåga att imitera manuella gester visade sig också ha samband med läsförmåga. Mer specifikt, så fanns det ett samband mellan precision i att imitera obekanta tecken och grad av utveckling i ordläsning (Artikel IV). Dessutom föreföll precision i att imitera välbekanta tecken. vara associerat med utveckling i läsförståelse (Artikel IV). Resultaten visade vidare att arbetsminne och Theory of Mind (ToM) är relaterade till läsförståelse hos D/H barn som använder sig av teckenspråk och är i början av sin läsutveckling (Paper III). Vid sidan av resultaten rörande läsutveckling, framkom också att teckenspråkserfarenhet ökar sannolikheten för etablering av representationer av manuella gester (Artikel II). Vidare uppvisade gruppen typisk progression i ToM, om än försenad (Paper III). Sammantaget ger resultaten från dessa studier stöd till uppfattningen att teckenspråkskunskaper kan vara en grund för läsutveckling hos D/H barn. En möjlig implikation av detta är att insatser som stöttar utveckling av teckenbaserade representationer och deras användning vid bearbetning av skrivet språk kan främja läsutveckling för D/H barn. Utifrån avhandlingens resultat föreslås en modell som beskriver vilka processer som ingår i bearbetning av skrivet språk. Enligt modellen fungerar arbetsminnet som en plattform för modalitetsöverskridande språkbearbetning och multimodal språkutveckling genom integration av inkommande stimuli och språkmedierade representationer.

List of Papers

Paper I

Holmer, E., Heimann, M., & Rudner, M. (2016). Evidence of an association between sign language phonological awareness and word reading in deaf and hard-of-hearing children. *Research in Developmental Disabilities*, *48*, 145-159. doi:10.1016/j.ridd.2015.10.008

Paper II

Holmer, E., Heimann, M., & Rudner, M. (2016). Imitation, sign language skill, and the Developmental Ease of Language Understanding (D-ELU) model. *Frontiers in Psychology*, *7*, 107. doi:10.3389/fpsyg.2016.00107

Paper III

Holmer, E., Heimann, M., & Rudner, M. (2016). Theory of Mind and reading comprehension in deaf and hard-of-hearing signing children. *Manuscript submitted for publication*.

Paper IV

Holmer, E., Heimann, M., & Rudner, M. (2016). Computerized sign language based literacy training for deaf and hard-of-hearing children. *Manuscript submitted for publication*.

Abbrevations

BSL British Sign Language
CHS Cognitive Hearing Science
CI Cochlear implant
C-PhAT-SSL Cross-modal Phonological Awareness Test, Swedish Sign Language version
C-PhAT-Swed Cross-modal Phonological Awareness Test, Swedish version
DHH Deaf and hard-of-hearing
DHH-AD Deaf and hard-of-hearing with additional disability
D-ELU Developmental Ease of Language Understanding
ELU Ease of Language Understanding
HA Hearing aid
HL Hearing loss
PA Phonological awareness
Pho Phonological
RCPM Raven's Coloured Progressive Matrices
RSNS Regional Special Needs Schools
Sem Semantic
SMS Swedish Manual Alphabet and Manual Numeral Systems
SSL Swedish Sign Language
ToM Theory of Mind
WC Wordchains
WM Working memory
WPRC Woodcock Passage Reading Comprehension

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Introduction

We live in an information society where career opportunities are often contingent on literacy, and the inability to read is associated with social exclusion. Children with hearing loss are at risk of delayed literacy, even when they use technical aids to assist hearing. Regional Special Needs Schools (RSNS) in Sweden are schools for deaf and hard-of-hearing children and have a bilingual curriculum in which sign language is used as a teaching medium. Just as in mainstream schools, acquisition of literacy skills is a major focus. Previous research has shown that among deaf signing children, sign language skill predicts reading ability and that some deaf signing children do become excellent readers. The work in the present thesis investigates the connection between sign language skill and literacy in RSNS pupils who are learning to read. It evaluates the effects of computerized sign language based literacy training and investigates concurrent and longitudinal associations between skills in the domains of sign language, cognition and reading.

Background

Disability and Hearing Loss

The bio-psycho-social model captured in the *International Classification of Functioning, Disability, and Health* by the World Health Organization (2001), describes disability as an ongoing interaction between the individual (biological level) and contextual factors, covering both environmental (social level) and personal (psychological level) aspects. A bio-psycho-social model and similar perspectives on disability (e.g., critical realism; Bhaskar & Danermark, 2006), acknowledges the importance of loss in bodily structure or function, and the social constraints that might restrict an individual's everyday functioning, as well as interactions between these levels and psychological factors. Indeed, individuals with exactly the same physical loss may vary in functional level due to mitigating or strengthening psychological and social factors. Deaf and hard-of-hearing (DHH) children are no exception. Individual characteristics and interventions at biological, psychological and social levels are all important factors for everyday functioning. The present work is framed within the bio-psycho-social model, and its primary focus is at a psychological level.

Cognitive Hearing Science

The bio-psycho-social model of disability advocates multidisciplinary research. Cognitive Hearing Science (CHS; Arlinger, Lunner, Lyxell, & Pichora-Fuller, 2009; Campbell, Rudner, & Rönnberg, 2009) is a multidisciplinary field that investigates interactions between cognitive factors and hearing ability (Rönnberg et al., 2013). One important outcome from this field of research, with theoretical, clinical and practical implications, is that specific psychological mechanisms can to some extent compensate for negative effects of hearing loss (HL). This idea is expressed in the Ease of Language Understanding (ELU) model (Rönnberg, 2003; Rönnberg, Rudner, Foo, & Lunner, 2008; Rönnberg et al., 2013), in which language understanding is described as being dependent on the interactions between the quality of the incoming language signal and both language specific and domain general cognitive mechanisms and representations. In particular, the

ELU model describes conditions under which language processing can become less or more challenging.

Deaf and Hard-of-Hearing Children in Sweden

Both in Sweden (Carlsson, 2005) and internationally (Mathers, Smith, & Concha, 2000), four categories of HL, relating to the degree of the loss, are commonly used for classification at the level of body function: mild HL, moderate HL, severe HL, and profound deafness. For the three first categories, hearing aids (HAs) may be used to amplify the sounds in the environment. In cases where HAs are not likely to produce any significant gains in speech perception, most typically profound HL, cochlear implants (CIs) may be considered (The Swedish Council on Technology Assessment in Health Care, 2006). CIs transmit a degraded signal, which nonetheless in many cases aids the establishment of functional levels of speech (Kral & Sharma, 2012; Nakeva von Mentzer, 2014; Löfkvist, 2014; Wass, 2009). However, speech outcome with technical aids always depends on factors not relating to the aids per se, such as the quality of language focused interventions or individual factors (Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007). Further, a technical aid never fully restores the quality of the spoken language signal, and any degree of HL may increase the risk of delayed speech development even when technical aids are used (Davis, Elfenbein, Schum, & Bentler, 1986; Moeller et al., 2007).

According to the Swedish Agency for Health Technology Assessment and Assessment of Social Services (2006), around 100 to 200 children are born each year in Sweden with a HL that requires some form of technical (e.g., CI) and/or communicative habilitation. A similar number of children also acquire HL before the start of formal schooling, e.g., due to accident or disease, and in total there are currently around 4500-5000 school aged DHH children in Sweden (Swedish Government Official Reports, 2011).

In the present work, the term deaf and hard-of-hearing (DHH) children is used to refer to all children who have a HL, and who in addition show atypical speech development and are furthermore in need of interventions that may be technical (e.g., CI) or communicative or both (for a similar definition, see Lederberg, Schick, & Spence, 2013). Some DHH children (5-10%, Kelly & Barac-Ciroja, 2007; Lederberg et al., 2013) grow up in an environment where they are exposed to a sign language from birth and these children show typical language development in the manual-visual modality instead of the oral-aural modality (Lederberg et al., 2013). Most DHH

children, however, do not show typical language development. This is due to restricted input of sign language and the degraded quality of the speech signal (Campbell, MacSweeney, & Woll, 2014; Lederberg et al., 2013). In Sweden, many DHH children use both sign language and spoken language, although they typically prefer one of these languages for communicative and learning situations (Svartholm, 2010). In line with the bilingual curriculum, DHH children who attend Regional Special Needs Schools (RSNS) in Sweden are taught in Swedish Sign Language (SSL) and in spoken and/or written Swedish (The National Agency for Special Needs Education and Schools, 2016).

Sign language

Sign languages are natural manual languages that are used by deaf people around the world, and which are functionally and structurally equivalent to spoken languages (for a review, see Emmorey, 2002). In contrast to gestures, sign languages are symbolic, conventionalized, and compositional (Corina & Knapp, 2006). Like spoken languages, sign languages have sub-lexical, lexical and syntactic structures (Emmorey, 2002). However, signs are performed manually and perceived visually whereas words are performed orally and perceived aurally. This means that the phonology, relating to the sub-lexical language structure, differs across language modality. In sign languages, the sub-lexical structure is defined based on the formation and movements of hands and arms, as well as place of articulation and non-manual features (e.g., facial expression) (Brentari, 2011), whereas in spoken languages, the sub-lexical structure consists of speech sounds, produced by forcing air through the vocal tract (Ladd, 2011). Today there are several hundred known sign languages around the world (Siegal, 2004), all of which are culturally specific and independent of the ambient spoken language (Corina, Gutierrez, & Grosvald, 2014). For example, in Sweden there is SSL, in Britain there is British Sign Language (BSL), and in the USA there is American Sign Language.

Regional Special Needs Schools

The first Swedish school for deaf children was founded early in the 19th century (Schönström, 2010). In the beginning, sign language was used for educating children at this school, and during the same century a standardized form of SSL started to emerge (Institute for Language and Folklore, 2014). However, in the 1860s, education for deaf children in Sweden began moving towards the oral method, that is, deaf children were supposed to learn to speak and lip-read for communication (Schönström, 2010). In 1880 the oral

method for deaf education was accepted internationally at a congress for teachers of deaf children held in Milan. As a consequence, sign language was forbidden in schools for the deaf. Thus, from 1880 deaf children in Sweden were taught in accordance with the oral method, and this view dominated deaf education for a century. Then, in 1981, SSL was officially acknowledged as the first language of deaf individuals in Sweden by the Swedish government. This acknowledgment was part of the movement towards a new educational curriculum for deaf children, in which SSL was defined as the first language for deaf children and Swedish was defined as a second language. Today, there are five RSNS in Sweden. These schools provide a learning environment that involves both SSL and written and/or spoken Swedish (Svartholm, 2010). For the present project, participants were recruited from these schools to ensure that SSL was used for communication and learning. However, it should be noted that there are DHH children in mainstream education who also use SSL (Holmström, 2013; Svartholm, 2010).

Of all DHH school aged children in Sweden today, only a small minority, today represented by 368 pupils (Swedish National Agency for Education, 2016), attend RSNS. Between the years 2005 and 2015 the number of pupils at these schools decreased by just below 30% (Swedish National Agency for Education, 2006; 2015). However, as can be seen in Figure 1, this attrition seem to be slowing down. The pattern indicates that over the last decade more DHH children were enrolled in mainstream education than earlier. This probably reflects the fact that nowadays in Sweden more children with profound deafness receive CI early in life and develop functional levels of speech. Indeed, during the same time period, the number of schoolchildren with profound deafness not using CI was reduced by around 90% (Hörselskadades Riksförbund, 2005; 2014).

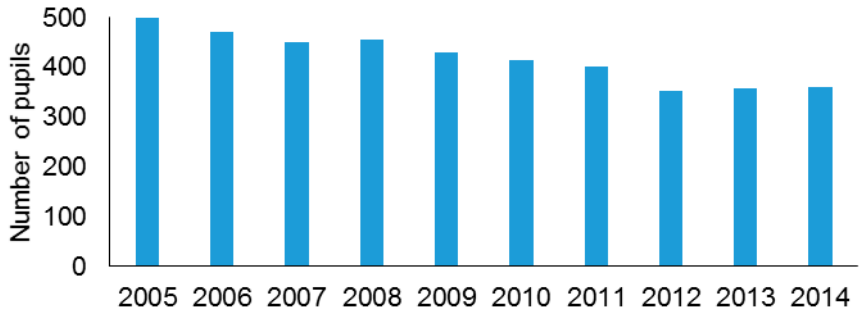

Figure 1. Pupils attending Regional Special Needs Schools.

RSNS pupils represent a heterogeneous group. Although many parents to DHH children in Sweden learn some SSL, there is great variability in how much sign language DHH children are exposed to (Svartholm, 2010). As mentioned above, only 5-10% of all DHH children are born to deaf parents who are native sign language users. Exposure to sign language at home is therefore highly variable between pupils. Further, some RSNS pupils use technical aids, and others do not. Further, some originate from another country and did not receive technical aids until they moved to Sweden, while others are born in Sweden and have been implanted early in life, but did not show the expected progress in speech development. Another group has additional medical or developmental conditions in combination with their HL. All use SSL in school, sometimes in combination with spoken Swedish. In summary, the prerequisites for learning are likely to differ between any two given pupils at these schools.

Language, Cognition, and Reading

Based on theoretical and empirical considerations three skills at the intersection between language processing and cognition were investigated in the present work. These skills were working memory, imitation and Theory of Mind (ToM), and the primary focus was on how they relate to developing reading skills in RSNS pupils.

Language processing

Language has its specific knowledge base, relating to the lexicon and syntax, as well as its unique functional outcomes, like efficient sharing of thoughts and ideas or creating stories and instructions (Hagoort & Levinson, 2014). In line with the literature (Hagoort & Indefrey, 2014; Hagoort & Levinson, 2014; Kintsch & Rawson, 2007; Perfetti & Stafura, 2014; Zwaan, 2015), word level processing, i.e., the processing and understanding of lexical items (e.g., words or signs), and language comprehension, i.e., understanding the meaning of one lexical item or several lexical items in a sequence (e.g., sentences), are herein assumed to represent connected albeit qualitatively different processes. Successful identification of lexical items as a language signal represents a starting level of language understanding. The basic function of lexical items is that they can be used by the perceiver to approximate the real world by connecting the words to their corresponding semantic representations, which refers to their meanings, or more generally, knowledge about the world and objects in it (Binder, Desai, Graves,

& Conant, 2009; Csibra & Shamsudheen, 2015). Thus, when a lexical item is successfully detected and its meaning can be accessed from long-term memory, it may be possible to understand the utterance. Furthermore, in longer messages involving several lexical items, the order in which the items are organized provides a structure for understanding how the meanings of those items are related to each other (Kintsch & Rawson, 2007). Producing rich and meaningful representational models from a language signal thus relies on language specific knowledge, domain general semantic knowledge and cognitive mechanisms, such as working memory, that support the integration of different sources of information and keeping a representational model in memory, and appropriate inference making (Kintsch & Rawson, 2007; Rönnberg et al., 2013; Zwaan, 2015). Further, language understanding is likely to become more precise if the intent of the speaker is taken into account (Hagoort & Indefrey, 2014), which involves ToM. Sign and spoken languages are processed in a similar manner at different linguistic levels, including the sub-lexical, lexical, and syntactic (for reviews, see Corina et al., 2014; MacSweeney, Capek, Campbell, & Woll, 2008). Efficient language processing of spoken (Rönnberg et al., 2013), sign (MacSweeney, Capek, et al., 2008), and written (Perfetti & Stafura, 2014) languages is thus likely to involve language specific knowledge, domain general knowledge and cognitive mechanisms.

Working memory and language processing

Working memory is the cognitive system which maintains items during a short period of time and processes them (Baddeley, 2012; Ma, Husain, & Bays, 2014). An item refers to a chunk with bits of information whose number in theory have no upper limit (Miller, 1956). Chunking is an important mechanism in short-term maintenance of items since it influences processing of incoming stimuli based on prior representations (Gobet et al., 2001).

It is of theoretical importance to make a distinction between short-term memory and working memory. The former is traditionally operationalized in simple span tasks as the number of items that can be maintained in memory during short intervals. The latter, on the other hand, is measured in complex span tasks in which items have to be remembered for a short period of time at the same time as the items are manipulated or storage is interfered with in some other way (Baddeley, 2012). In adults, simple span for items that can be represented verbally is 7 +/- 2 items (Miller, 1956), whereas simple span for

items that cannot easily be represented verbally is lower, around 3-5 items (Cowan, 2000). For items of linguistic nature in particular, there is also a developmental progression in short-term maintenance of items (Diamond, 2013). For deaf signers, simple span for signs is closer to simple span for items that cannot be represented verbally (e.g., Boutla, Supalla, Newport, & Bavelier, 2004). This applies even when pre-existing representations of those signs are available in the mental lexicon (Wilson & Emmorey, 1998). The reason for this are not completely understood and there are several competing explanations (c.f., Andin et al., 2013). On the other hand, complex span for signs and words is similar (Andin et al., 2013; Boutla et al., 2004). Further, brain imaging studies indicate that similar structures are involved in working memory tasks for signers and non-signers (Rudner, Andin, & Rönnberg, 2009), which suggests that similar cognitive components are involved.

Working memory is involved in any process that unfolds over time, connecting what just happened to what is about to happen (Diamond, 2013), and is a key component of language processing (Baddeley, 2012; Rönnberg et al., 2013). Language processing does not always put pressure on the limits of working memory capacity. For example, using a small set of familiar words to construct and maintain a model of a representation of a familiar event is an effortless process. However, when the number of words increases, familiarity with the words decreases, or contextual demands (e.g., background noise) interfere with processing, the load on working memory increases and remembering becomes challenging (Ma et al., 2014; Rönnberg et al., 2013). Resource models of working memory (Ma et al., 2014) explain this by suggesting that the interaction between load, that is, the amount of information to be remembered, and the distinctiveness of the items, that is, how precisely the information can be represented, is key in short-term maintenance of items. Children who are still learning to read struggle to find phonological and semantic representations that match written words. The process of finding appropriate representations taxes working memory capacity. Thus, working memory capacity might be involved in the process of reading, especially before full literacy skills have been established. Working memory has indeed been linked to both word reading and reading comprehension in children who are an early stage of their reading development (National Institute for Literacy, 2008).

The Ease of Language Understanding model

The ELU model (Rönnberg, 2003; Rönnberg et al., 2008; Rönnberg et al., 2013) suggests that language processing builds on meaning prediction, and the model describes how language understanding takes place under more or less demanding conditions for language processing (see Figure 2). When the incoming language signal is distinct and familiar, the ELU model predicts that language processing will be efficient and easy. The incoming language signal is matched to a stored representation in a process that involves the Rapid, Automatic, and Multimodal Binding of PHOnology (RAMBPHO). If the number of phonological attributes reach a certain threshold, it is easy to *Match* the representation to a stored lexical representation (see Figure 2). However, language understanding can become difficult due to, e.g., sensory impairments, interference from background noise, distortion of the language signal, or competing lexical candidates all of which partially match the incoming signal. When language understanding becomes difficult, the ELU model predicts a qualitative change in the characteristics of language processing. In particular, the language signal enters an *Explicit processing loop* (see Figure 2), which is constrained by working memory capacity (WM in Figure 2), and long-term representations (LTM in Figure 2) are invoked, e.g., by inference, to aid understanding.

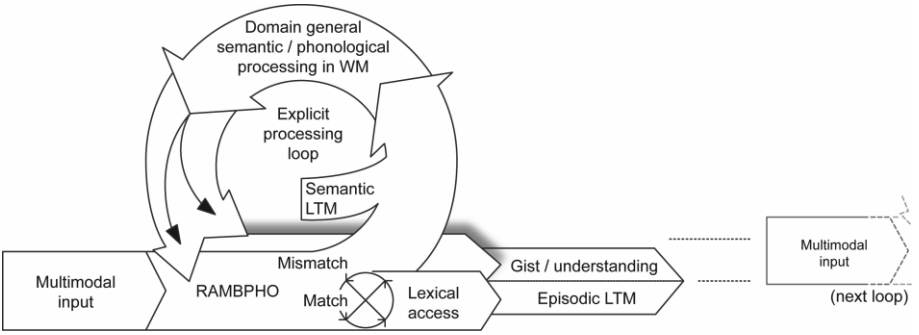

Figure 2. The Ease of Language Understanding (ELU) model. Reprinted from "The Ease of Language Understanding (ELU) model: Theoretical, empirical, and clinical advances" (Rönnberg et al., 2013). Copyright 2013 by Rönnberg et al. under the CC BY 3.0 license.

A child who is learning to read may well know the meaning of the words in the text, but be less able to connect prior representations to their written form. In relation to the ELU model (Rönnberg et al., 2013), reading reflects a

demanding language condition for the learning child and is likely to be constrained by working memory capacity as well as availability of long-term representations (c.f., Perfetti & Stafura, 2014).

Imitation

For the primary group of interest in the present study, i.e., DHH children who use sign language, manual gestures are sometimes signs, and for these children imitation of gestures might thus represents a special act. Imitation, or the repetition of a behavior of another person (Brass & Heyes, 2005), is regarded as a key mechanism in cognitive development for all children (Heyes, 2016; Meltzoff & Williamson, 2013; Tomasello, Kruger, & Ratner, 1993). Even though imitation is relatively easy to define at the surface level, there are different views on which cognitive mechanisms are involved in connecting what is perceived (e.g., visually or auditory) to the execution of a similar behavior. The key question is how perception is transferred into behavior (Heyes, 2016; Iacoboni, 2009). Within transformational theories on imitation (Heyes, 2016), perception is assumed to be converted into an executory plan via a domain general representation that is neither a motor nor a perceptual representation (Meltzoff & Moore, 1997). Associative theories, on the other hand, suggest that imitation is the result of a direct mapping between perceptual input and motor output (Heyes & Ray, 2000; Heyes, 2001; 2016). Input and output are thus viewed as two sides of the same coin. However, it is possible that imitation relies on both domain general processes (Meltzoff & Moore, 1997) and prior representations (Heyes & Ray, 2000).

Efficient language processing and imitation

Even though imitation is not explicitly described as a part of the ELU model (Rönnberg et al., 2013), in the present work the model was used to derive theoretically driven predictions for the imitation of linguistic and non-linguistic materials. The ELU model proposes that the quality of an incoming language signal influences the processing of the signal. Basically, the greater the overlap between the signal and prior representations, the greater the likelihood of efficient processing. For the present work, it was assumed that the precision of imitation is a marker of processing efficiency. Thus, it was derived from the ELU model that imitation of familiar lexical forms, invoking both semantic and phonological information, is likely to be more precise than imitation of unfamiliar lexical forms, invoking only

phonological information, which in turn is likely to be more precise than imitation of behaviors that are non-lexical, that is, signals that show only weak overlap with lexical representations. The same expectations can also be derived from associative accounts (Heyes & Ray, 2000) of imitation. Thus, by manipulating the type of manual gestures to imitate (e.g., lexical or non-lexical), imitation of manual gestures can be used as a way to investigate processing of prior representations in sign language users (c.f., Marshall, 2014).

For hearing children, more precise repetition of familiar words in comparison to unfamiliar words, that is, legal lexical forms that have no meaning, has been reported (Casalini et al., 2007; Dispaldro, Deevy, Altoé, Benelli, & Leonard, 2011; Roy & Chiat, 2004; Sundström, Samuelsson, & Lyxell, 2014), and deaf children have been reported to be more likely than hearing non-signing children to imitate unfamiliar signs correctly (Mann, Marshall, Mason, & Morgan, 2010). Brain imaging studies also indicate that when deaf adults process lexical manual gestures, the activity in language related brain regions is greater than it is for hearing individuals (Cardin, Orfanidou, Rönnberg, Capek, Rudner, & Woll, 2013; Li, Xia, Zhao, & Qi, 2014; Newman, Supalla, Fernandez, Newport, & Bavelier, 2015). These findings demonstrate that the processing of manual gestures changes with sign language experience. On the other hand, there are also brain imaging studies indicating that lexical manual gestures are processed more efficiently than non-lexicalized manual gestures both by signing and non-signing individuals (e.g., Cardin et al., 2016). Thus, regardless of knowledge of sign language, imitation of lexical in comparison to non-lexical manual gestures might be easier, possibly reflecting that domain general representational processes also influence imitation (Meltzoff & Moore, 1997). However, the precision of imitation of familiar signs, unfamiliar signs and non-signs, that is, manual gestures that lack resemblance with lexical signs, has not yet been investigated across signing and non-signing children.

Although imitation and reading appear to be dissimilar at a surface level, the underlying cognitive mechanisms may sometimes be quite similar. In particular, tasks involving imitation of utterances might reflect cognitive processing of language that is important for developing reading skills. For example, the repetition (i.e., imitation) of lexical forms has been suggested to reveal individual differences in the ability to access, temporarily store and manipulate lexical information, both for spoken language users (Gathercole, 2006) and sign language users (Marshall, 2014). In particular, tasks involving

imitation of unfamiliar lexical forms may indicate how susceptible the lexical system is to change (e.g., Metsala, 1999; Gathercole, 2006). Further, imitation of unfamiliar lexical forms is linked to reading skills in hearing children (Melby-Lervåg & Lervåg, 2012; Pennington & Bishop, 2009) and associated with the ability to read words in DHH children who use speech (Dillon & Pisoni, 2006; Nakeva von Mentzer et al., 2015). Imitation of lexical manual gestures may reveal the ability of sign language users to manipulate representations from the lexical system, and may thus involve processes of importance for reading development. Prior to the present work, however, no one had investigated how the ability to imitate different types of lexical manual gestures relates to developing reading in DHH signing children, although imitation of sign language has been connected to reading skills in earlier studies of deaf adults (e.g., Freel, Clark, Anderson, Gilbert, Musyoka, & Hauser, 2011; Stone, Kartheiser, Hauser, Petitto, & Allen, 2015).

Theory of Mind, sign language skill and reading

ToM is the ability to represent processes and states of mind in oneself and others, and to explain behavior in terms of mental states (Frith & Frith, 2012). In the early studies on ToM in the developmental literature, false belief tasks were introduced (Wimmer & Perner, 1983) as a way to investigate whether children had or had not attained an ability to understand the mental states of others. This approach reflects a view on ToM as an all-or-nothing capacity (Baron-Cohen, Leslie, & Frith, 1985) that is in place in typically developing children around the age of four as indicated by their ability to solve false belief tasks (for a meta-analysis, see Wellman, Cross, & Watson, 2001). However, reducing ToM to a score on false belief tasks has been criticized for a number of reasons. In particular, it has been pointed out that the complexity of the task taxes language comprehension skills and is likely to load on working memory (Bloom & German, 2000). Further, false belief is only one part of the construct of ToM (e.g., Flavell, 2004; Reddy, 2008). Accordingly, attempts have been made to investigate ToM using tasks other than false belief (e.g., Baillargeon, Scott, & He, 2010; Dziobek et al., 2006; Wellman & Liu, 2004). For example, Wellman and Liu's ToM scale assess the ability to reason on how desires, beliefs, knowledge access, and false beliefs influence behavior, as well as the ability to understand that a person can express one emotion but experience another.

Development of Theory of Mind

Before children solve false belief tasks, they actually do seem to understand other aspects of mental operations. For example, at the age of two, children seem to understand desires as a cause of behavior, and at the age of three, they start to differentiate between their own beliefs and knowledge and those of others (Carlson, Koenig, & Harms, 2013; Wellman, 2014). Further, an understanding of sarcasm and the fact that people sometimes hide their true feelings seems to develop after the age of four (Wellman, 2014). Even though similar findings relating to mind reading (Heyes & Frith, 2014) during the pre-school years have been reported from several cultural settings (Wellman, 2014), there also seems to be some cultural diversity (Heyes & Frith, 2014; Wellman, 2014) which suggests that ambient culture influences how children learn to read minds. Also, there is currently much debate concerning whether or not evidence of ToM can be observed during the first year of life (e.g., Baillargeon, Scott, & Bian, 2016; Perner, 2014; Ruffman, 2014). For example, during the first year of life, infants react to violations related to other individuals' goal directed behaviors and preferences, and they seem to be able to predict actions (for a review, see Baillargeon et al., 2016).

DHH children often display delayed development of ToM (Lederberg et al., 2013; Peterson, 2009; Sundqvist & Heimann, 2014; Wellman, 2014). Despite these delays, the progression of developmental achievements does seem to be similar across normally-hearing and DHH children (Peterson, O'Reilly, & Wellman, 2016; Peterson, Wellman, & Liu, 2005; Peterson, Wellman, & Slaughter, 2012). In fact, studies on DHH signing children have provided some important insights into the way in which early interaction between children and their caregivers supports ToM development. Most prominently, among DHH children, those who have deaf signing parents display typical ToM development, whereas those who do not have signing parents typically show delays in this domain (Lederberg et al., 2013; Peterson, 2009). Further, there is also evidence that age of implantation of CI is associated with ToM development in DHH children who have hearing parents and use spoken language (e.g., Sundqvist, Lyxell, Jönsson, & Heimann, 2014). Findings like these, indicate that the establishment of a functional language during the early years of life is important for later ToM development. Accordingly, both language development and rich language interactions early in life, in particular discussion of mental states like desires and beliefs (Astington & Dack, 2008), have been suggested to be important for ToM development (e.g., Lederberg et al., 2013; Milligan, Astington,

& Dack, 2007; Peterson, 2009; Sundqvist & Heimann, 2014; Wellman, 2014). Studies indicate that the way parents use mental state language in interaction with their child during the first year of life predicts the child's later development in ToM (e.g., Kirk, Pine, Wheatley, Howlett, Schulz, & Fletcher, 2015; Meins, Fernyhough, Wainwright, Das Gupta, Fradley, & Tuckey, 2002; Sundqvist, Koch, Holmer, & Heimann, 2014). Further, even though ToM is a unique cognitive construct, it has at the same time been suggested to rely on general language development (Milligan et al., 2007), certain linguistic accomplishments (de Villiers & de Villiers, 2014) and working memory (Carlson et al., 2013; Moses & Tahiroglu, 2010). Siegal and Varley (2002) suggest that ToM development is supported by co-opted systems, including language and working memory, and is triggered by early conversational experiences of mental states. In the present thesis, ToM is viewed as a multifaceted construct, involving specific representations of mental states, knowledge about how these relate to behavior, and mechanisms supporting the application of ToM. The Wellman and Liu ToM scale (2004) was used in the work presented in this thesis to capture a broader conceptualization of ToM than that represented by false belief. However, it should be noted that even the ToM scale is not exhaustive (c.f., Baillargeon et al., 2016; Reddy, 2008; Wellman, 2014).

Reading ability and Theory of Mind

Most research on ToM has been focused on its development and anomalies. However, individual variability in ToM also seems to influence learning processes (Carlson et al., 2013; Kloo & Perner, 2008). It is of particular interest in the present work that ToM has been linked to reading skills in hearing children (Blair & Razza, 2007; Kim, 2015a; Lecce, Caputi, & Hughes, 2011; Lecce, Caputi, & Pagnin, 2014; Miller, Müller, Giesbrecht, Carpendale, & Kerns, 2013; Ricketts, Jones, Happé, & Charman, 2013; Åsberg, Kopp, Berg-Kelly, & Gillberg, 2008). Specifically, ToM seems to be connected to reading comprehension (e.g., Kim, 2015a), but perhaps not to lower level reading skills (e.g., Miller et al., 2013), after controlling for other relevant variables like general language skills and working memory. Marschark and Wauters (2011) stated that ToM is "… relevant to academic performance and reading in particular…" (p. 495) for deaf children. Further, Courtin, Melot and Corroyer (2008) suggested that a more advanced ability to understand and reflect upon mental states indicates a capacity to go beyond what is perceptually present. This, in turn, may be of particular importance

for any child to become a proficient reader. Prior to the present work, connections between ToM and developing reading skills had not been investigated in DHH signing children. The empirical findings linking ToM to reading comprehension in hearing children (e.g., Kim, 2015a), as well as reports on difficulties within this domain in the general population of DHH children (Lederberg et al., 2013; Peterson, 2009), motivated the investigation of an association between ToM and reading comprehension in the present work.

Reading Development in Deaf and Hard-of-Hearing Signing Children

In an North American context, 5% of profoundly deaf individuals become proficient readers (Grade 12 equivalent; Kelly & Barac-Ciroja, 2007), and it has been estimated that the median level of reading for DHH individuals at the end of secondary education corresponds to a Grade 3 to Grade 4 hearing reader (Qi & Mitchell, 2012). In a European context, Rudner, Orfanidou, Cardin, Capek, and Rönnberg (2012) reported a mean level of reading corresponding to Grade 11 (i.e., reading age, 16 years) in a group of adult deaf native users of BSL. Delays in reading development have also been reported in a Swedish context (e.g., Heiling, 1994; Petersson, Liljestrand, Turesson-Morais, Eriksson, & Hendar, 2000). However, Heiling (1994) reported that close to 50% of a group of Grade 8 RSNS pupils performed as well or better than Grade 4 hearing children on a standardized reading task. Although these numbers demonstrate that reading is a difficult hurdle to tackle for DHH children, they also indicate variability and that some become proficient readers. For DHH children, the difficulty in learning to read has to do in part with the fact that access to speech sounds is limited. Sign-based representations at the sub-lexical and syntactic levels cannot be directly mapped onto the structure of speech-based language as manifested in written text. Thus, for DHH signing children the link between sub-lexical representations and text is either poorly specified or indirect.

For DHH children both the quality and quantity of language input in early years is often limited, and this is likely to influence later cognitive and language development, including reading (Lederberg et al., 2013; Mayberry, Chen, Witcher, & Klein, 2011; Mayberry, 2007). More specifically, the early language environment influences how well DHH signing children can establish language skills that they can utilize to learn to read

(Goldin-Meadow & Mayberry, 2001; Hoffmeister & Caldwell-Harris, 2014; Mayberry, 2007). It has been suggested that strong sign language skills can provide a base for reading development (e.g., Hoffmeister & Caldwell-Harris, 2014). Scientists who emphasize the importance of strong sign language skills suggest that sign-based semantic representations can become associated with written words and that from this the meaning of the written words can be extracted (Crume, 2013; Hermans, Knoors, Ormel & Verhoeven, 2008a; Haptonstall-Nykaza & Schick, 2007; Hoffmeister & Caldwell-Harris, 2014). Further, sign language skills and sign-based representations can be used to reflect upon, discuss and elaborate on the content of text together with others who have already mastered reading (Hoffmeister & Caldwell-Harris, 2014; Mayberry, 2007; Svartholm, 2010). It is possible that these activities support effective comprehension strategies and understanding of second language structures. Sign language ability may also mark more general language skills, e.g., breadth and depth of vocabulary or the ability to establish and use representations that can support reading development (Goldin-Meadow & Mayberry, 2001; Hoffmeister & Caldwell-Harris, 2014; Mayberry, 2007). Better understanding of how reading works for DHH signing children may both have important theoretical and practical implications. Similarities and differences across sign language and speech users can help us identify key language and cognitive mechanisms of reading. Further, it can also inform us on how to intervene with delayed reading development and how to construct educational policies.

Word reading and reading comprehension

In the present work, reading is viewed as a multi-componential process that involves language general and language specific knowledge, as well as cognitive skills that support precise and enduring models of events and objects in the text (Hulme & Snowling, 2014; Kamhi & Catts, 2012; Perfetti & Stafura, 2014). It is well established that word reading and reading comprehension are connected, both in hearing (Garcia & Cain, 2014; Ripoll Salceda, Alonso, & Castilla-Earls, 2014) and deaf children (Marschark & Wauters, 2008). However, as outlined in the section on cognition and language processing, lexical processing (e.g., word reading) and language comprehension are not completely interchangeable, and this also applies to written language (Perfetti & Stafura, 2014). In the typical case, word reading seems to rely on phonological representation and analysis, while reading

comprehension relies on semantic analysis, integration of different sources of knowledge, appropriate inference making, and syntactic knowledge.

Sign language skills and learning to read words

Typically, the first stage of learning to read involves correctly identifying written words. This process can either be done by matching a written word, i.e., orthographic form, to stored phonological representations (Stanovich, 1982), or by directly connecting the written word to its meaning (Coltheart, 2006). For children who use speech, a phonological route involves learning to transform individual letters into their phonological equivalents, which requires stored representations of phonemes and the ability to assemble phonemes into words (Ziegler & Goswami, 2005). When orthography is recoded into a phonological form, the spoken word may or may not correspond to a stored lexical item. When it does, meaning can be accessed. When it does not, a new word might be learned, given that the meaning of the word can be inferred from the context or is provided by a more knowledgeable reader. The Neighborhood Activation Model of language processing proposes that the incoming language signal is analyzed in steps (Luce & Pisoni, 1998). The onset of a word which involves its initial phonological unit (Ahlsén, 2006) activates the group of stored lexical items that begin with that unit, of which one may be the target word (Luce & Pisoni, 1998). Then, as the language signal unfolds sound by sound, a restricted number of candidates will be left, and as the rime, that is, the sounds following the onset (Ahlsén, 2006), ebbs out, a single lexical candidate is selected (Luce & Pisoni, 1998). The process of mapping an orthographic form to stored phonological representations is called decoding (Stanovich, 1982), and is a key to learning to read. However, some suggest that written words can also be directly mapped to their meaning, bypassing phonology via an orthographic route (Coltheart, 2006). Whether or not orthographic forms can be stored as lexical items is the subject of debate (Leinenger, 2014). This is, however, not investigated in detail in the present work. Even though current evidence does not rule out the involvement of orthographic processes in reading (Bélanger & Rayner, 2015; Grainger, Dufau, & Ziegler, 2016), it suggests that the route from written words to meaning always involves some kind of phonological code (for a review, see Leinenger, 2014). The term word reading or word identification is used in the present work when referring to the process of reading words accurately,

regardless of the degree to which phonological, orthographic, and semantic processes are involved.

Even though not identical at the perceptual and functional levels, spoken language and alphabetic written language correspond at the sub-lexical, lexical and syntactic levels (Kamhi & Catts, 2012). Thus, there is a close structural mapping between speech and written language. However, because speech representations are likely to be weak in most DHH children due to degraded speech input, it is unlikely that speech based representations support text analysis, even though there are exceptions to this rule (e.g., Kyle, Campbell, & MacSweeney, 2015). On the other hand, DHH signing children have sign-based representations which may support development of word reading skills. Indeed, experimental evidence indicates that there is a sign-based route to word reading for DHH signers (Barca, Pezzulo, Castrataro, Rinaldi, & Caselli, 2013; Conlin & Paivio, 1975; Kubus, Villwock, Morford, & Rathman, 2015; Morford, Kroll, Piñar, & Wilkinson, 2014; Morford, Wilkinson, Villwock, Piñar, & Kroll, 2011; Ormel, Hermans, Knoors, & Verhoeven, 2012; Pan, Shu, Wang, & Yan, 2015; Treiman & Hirsh-Pasek, 1983). This is quite remarkable given that sign language and written language do not correspond in sub-lexical structure. For example, Morford et al. (2011) reported that semantic similarity judgements were faster when written words had phonologically similar sign translations than when they were phonologically unrelated. Similar findings were reported in the case of deaf children by Ormel et al. (2012). Although these findings indicate that sign phonology support word identification in deaf signers, response times, often measured at the level of seconds, are rough measures of lexical retrieval, since the retrieval process takes only a few hundred milliseconds (Leinenger, 2014). As a stronger demonstration of an early effect of sign phonology in word reading, in an eye-tracking study in which participants read sentences, Pan et al. (2015) reported a preview cost of words presented parafoveally, i.e., within a 2° to 5° visual angle from a fixation point (Schotter, Angele, & Rayner, 2012). This cost was apparent when the sign language equivalent had a phonological overlap with the target word but not when words presented parafoveally were unrelated in terms of sign phonology (Pan et al., 2015). Pan et al.'s (2015) findings may thus indicate that sign phonology is involved in word reading in DHH signing individuals. Taken together, the empirical findings reviewed above suggest that DHH signers may recode print into a sign-based code when they are learning to read. Thus,

strengthening the connection across language modalities might improve reading development in DHH signing children.

The mapping between orthographic forms and stored phonological representations may be supported by similar cognitive mechanisms across users of speech and sign language. Indeed, there is growing consensus that the cognitive mechanisms involved in word reading are likely to be similar for hearing and DHH children, even though the mechanisms may have become specialized in different modalities due to different language experiences (Andrews & Wang, 2015). Phonological processing skills influence reading development in hearing children (Melby-Lervåg, Lyster, & Hulme, 2012; National Institute for Literacy, 2008; Wagner & Torgesen, 1987). In particular, sensitivity to sub-lexical structure, or phonological awareness (PA), is related to word reading in hearing children (Melby-Lervåg et al., 2012) and DHH children who primarily use speech (e.g., Colin, Magnan, Ecalle, & Leybaert, 2007). PA has been suggested to reveal efficient access to phonological representations (e.g., Melby-Lervåg et al., 2012; Wagner & Torgesen, 1987), and may thus reflect a domain general processing mechanism (c.f., MacSweeney, Waters, Brammer, Woll, & Goswami, 2008) that is important for reading development in all children. Indirect support of this notion comes from studies indicating similarities in sub-lexical and lexical processing across the spoken and manual modality at a neural (Cardin et al., 2016; Gutiérrez, Müller, Baus, & Carreiras, 2012; Hosemann, Herrmann, Steinbach, Bornkessel-Schlesewsky, & Schlesewsky, 2013; MacSweeney, Waters, et al., 2008) as well as a behavioral level (Baus, Gutiérrez, & Carreiras, 2014; Berent, Dupuis, & Brentari, 2014; Carreiras, Gutiérrez-Sigut, Baquero, & Corina, 2008; Corina, Hafer, & Welch, 2014). However, prior to the present work, only one study had investigated associations between sign language PA and reading in DHH signing children, indicating a positive relationship (McQuarrie & Abbott, 2013). Further, as mentioned above, imitation of unfamiliar lexical forms, involving manipulation of stored phonological representations (Gathercole, 2006; Marshall, 2014), has been linked to reading skills in both hearing (Melby-Lervåg & Lervåg, 2012; Pennington & Bishop, 2014) and DHH children (Dillon & Pisoni, 2006; Nakeva von Mentzer et al., 2015), and might thus play an important part in word reading development in DHH signing children. The relations between phonological analysis and processing of sign language and developing word reading skills were of particular interest in the present work. However, working memory might constrain phonological

processing (Gathercole, 2006; Rönnberg et al., 2013) and is connected to word reading in both hearing (National Institute for Literacy, 2008) and DHH children (e.g., Daza, Phillips-Silver, Ruiz-Cuadra, & López-López, 2014; Kyle & Harris, 2010); thus, the relation between working memory capacity and word reading was also investigated in the present work.

Although there is no one-to-one mapping between signed and written languages at sub-lexical and syntactic levels, written letters and digits can be manually represented with manual alphabets and manual numeral systems (Bergman & Wikström, 1981; Padden & Gunsauls, 2003). In these systems, letters and digits are represented by specific handshapes, sometimes involving a movement. Using these symbols to represent a written word in the manual modality is referred to as fingerspelling (Morere & Roberts, 2012). It has been suggested that fingerspelling can actually provide sign language users with a phonological code of written words in the manual modality that they can use as a bridge across language modalities (Crume, 2013; Haptonstall-Nykaza & Schick, 2007), and there is some empirical support for this (Hanson, Liberman, & Shankweiler, 1984; Haptonstall-Nykaza & Schick, 2007; Stone et al., 2015). In particular, it has been suggested that fingerspelling may support the development of precise and rapid word identification (Stone et al., 2015). In Sweden, fingerspelling is commonly used (Roos, 2013) and some fingerspelled words are actually lexicalized in SSL (for further discussion, see Andin, Rönnberg, & Rudner, 2014). In the present work, connections between fingerspelling and reading skills were not addressed per se, but the characteristics of the Swedish manual alphabet and manual numeral systems were utilized to measure sign language PA.

Learning to comprehend texts as a sign language user

The ultimate goal of learning to read is to understand texts, or as (Chall, 1989) puts it: "…reading to learn…" (p. 28). Words and word order in a text provides an embedded message, but it is up to the reader to mentally construct an accurate model of the content of that message (Kamhi & Catts, 2012; Kintsch & Rawson, 2007; Perfetti & Stafura, 2014). That is, as in the case of general language processing, the language signal has to be transformed from a physical entity into a meaningful representation. Besides providing the reader with information, this also connects readers to the writer, who constructed the text in a specific way to communicate a certain message or story (Mar & Oatley, 2008; Zunshine, 2006), and to their cultural

context in a new way (Heyes & Frith, 2014). In the reading process, words establish the content of a text (Perfetti & Stafura, 2014) and, especially at the early stages of reading development, word reading is closely connected to reading comprehension in hearing children (Garcia & Cain, 2014; Ripoll Salceda et al., 2014), and an association also exists in deaf children (for a review, see Marschark & Wauters, 2008). However, as reading develops, typically there is a gradual shift towards heavier reliance on general language skills (Ripoll Salceda et al., 2014). Importantly, word reading and reading comprehension are connected but at the same time qualitatively different processes.

One of the most influential component models of reading comprehension is the Simple View of Reading (Gough & Tunmer, 1986; Hoover & Gough, 1990). The name of the model hints at its simplicity: the model only includes two components; a word decoding component, and a language comprehension component. Specifically, reading comprehension is described as the product of word decoding and language comprehension abilities. Thus, the absence of an ability either to decode words or to comprehend language leads to absence of reading comprehension. Further, a weakness in either of these skills could lead to difficulties in reading comprehension (Gough & Tunmer, 1986). The empirical support for the Simple View in typically developing children is extensive (Catts, Herrera, Nielsen, & Bridges, 2015; Hulme & Snowling, 2014; Language and Reading Research Consortium, 2015; Ripoll Salceda et al., 2014), and the model has also been suggested to be useful as a theoretical framework for reading in deaf children (Chamberlain & Mayberry, 2000; Stone et al., 2015). At the same time, it has been suggested that more detailed specification of the cognitive mechanisms involved in the two components of the Simple View would promote greater understanding of reading development (Hulme & Snowling, 2014; Kirby & Savage, 2008; Stuart, Stainthorp, & Snowling, 2008), and attempts have been made to achieve this (e.g., Byrne et al., 2013; Kim & Phillips, 2014; Kim, 2015a, 2015b; Tunmer & Chapman, 2012; Vellutino, Tunmer, Jaccard, & Chen, 2007).

We know less about reading comprehension in DHH signing children than we know about the word reading skills of this group (Kyle & Cain, 2015; Marschark & Wauters, 2008; Trezek, Wang, & Paul, 2011). This is partly because the literature on DHH signing children uses varying definitions of reading (Andrews & Wang, 2015). However, there are a couple of studies that have investigated reading comprehension that provide some

insights into which mechanisms that might be of particular importance when DHH signing children learn to understand written language. For example, Hermans, Knoors, Ormel, and Verhoeven (2008b) reported a positive association between sign vocabulary and written vocabulary in a group of deaf children. Further, Kyle et al. (2015) reported that vocabulary, either sign-based or speech-based depending on the preference of the participant, predicted unique variance in reading comprehension after controlling for other relevant variables (e.g., non-verbal cognitive ability) in a group of DHH children of whom approximately half used sign language. Results from another study indicate that vocabulary is also related to reading comprehension longitudinally in the same type of population (Kyle & Harris, 2010). The findings reviewed here indicate that regardless of modality, vocabulary may be a component of particular importance for developing reading comprehension in DHH children who use sign language. Further, in deaf signing adults, working memory has been reported to predict reading comprehension (Garrison, Long, & Dowaliby, 1997; Hirshorn, William, Hauser, Supalla, & Bavelier, 2015). In particular, Hirshorn et al. (2015) suggested that the maintenance of semantic information in working memory is the key to reading comprehension in deaf signing individuals. They suggested that working memory may compensate for weak speech based representations during comprehension of written language and support understanding of the syntactic rules of a second language. This notion is well in line with the ELU model (Rönnberg et al., 2013), which suggests that explicit processing of domain general semantic representations aids language understanding during challenging language conditions. There are also several studies indicating a positive relationship between general sign language skills and comprehension of written texts (Chamberlain & Mayberry, 2008; Freel et al., 2011; Heiling, 1994; Hoffmeister, 2000; Schönström, 2010) or general literacy skills (Strong & Prinz, 1997). The overall picture indicates that general sign language skills, in particular, maintenance of semantic representations in working memory, is of importance in the development of reading comprehension in DHH signing children. However, in general, studies have not focused on the specific connections between sign language skills and reading comprehension in DHH signing children who are learning to read.

Using interventions to support reading development

It is clear that sign language skills are involved in reading in DHH signing children. However, despite the long history of theoretical debate and empirical investigations, few have implemented interventions to support the connections between sign language skills and reading (for reviews, see Luckner & Cooke, 2010; Luckner & Handley, 2008; Luckner, Sebald, Cooney, Young, & Muir, 2005; Tucci, Trussell, & Easterbrooks, 2014). Some previous interventions have targeted aspects such as reading and comprehension strategies, inference making and background knowledge (e.g., Akamatsu & Armour, 1987; Walter, Munro, & Richards, 1998; van Staden, 2013). Others have implemented more specific interventions focusing on establishing connections between signs and written words (e.g., Reitsma, 2009; Wauters, Knoors, Vervloed, & Aarnoutse, 2001). In a study by Reitsma (2009), a group of deaf children worked on computer-based exercises in which they were tasked with learning the meaning of written words. Before training commenced, it was checked that they did not know the meaning of the words. The training involved four types of exercises. In two of the exercises, written words were paired with either a sign equivalent or a drawing depicting the meaning of the word. In the two other conditions, signs or drawings were matched to a written word, that is, meaning was matched to orthographic form. Post-training scores indicated improvements in both sign to word mapping and spelling of the words. In another study, Wauters et al. (2001) reported evidence indicating that providing DHH signing children with sign equivalents of written words may support development of accurate word reading. Hence, in line with theoretical notions (e.g., Hoffmeister & Caldwell-Harris, 2014), interventions aimed at establishing connections between written words and sign-based representations may be an effective way of supporting the development of connections between signs and written words in DHH signing children. However, whether such training also led to generalized improvement in word reading or reading comprehension has hitherto not been investigated.

Omega-is (Heimann, Lundälv, Tjus, & Nelson, 2004) is a top-down computerized literacy intervention, in which the user can explore written language in an interactive and stimulating environment. The theory underlying the program, Rare Event Transactional Theory (Nelson, 1998), suggests that the establishment of a new representation is a rare event, and that several contextual factors influence this process. For example, it is assumed to be influenced by how successfully the environment can trigger

activation of prior representations and help direct attention to the learning material. Individual factors, like emotional and cognitive resources and the richness of prior representations, are also deemed to be important. Computer-based interventions may be particularly well suited for supporting reading development in children for whom reading is difficult (Nelson, Welsh, Camarata, Tjus, & Heimann, 2001). In particular, it is possible to incorporate multiple associated representations to support comprehension, i.e., utterances as well as pictorial and video material, in a stimulating context. This learning environment supports the child's access to long-term representations, short-term maintenance and manipulation, sustained attention and engagement in working with the material, and is thus likely to facilitate learning (Mayer, 2008). In fact, Omega-is and its predecessors were developed with difficult learning situations in mind, and the program and its forerunners have shown positive effects on both word reading and reading comprehension in typically developing as well as children with, e.g., dyslexia, autism, and attention deficit hyperactivity disorder (e.g., Fälth, Gustafson, Tjus, Heimann, & Svensson, 2013; Gustafson, Fälth, Svensson, Tjus, & Heimann, 2011; Heimann, Nelson, Gillberg, & Karnevik, 1993; Heimann, Nelson, Tjus, & Gillberg, 1995; Helland, Tjus, Hovden, Ofte, & Heimann, 2011; Tjus, Heimann, & Nelson, 1998, 2004). Early versions of the program also indicated positive effects on reading development in deaf children (Prinz, Nelson, & Stedt, 1982; Prinz & Nelson, 1985). Thus, the Omega-is is likely to be a useful platform for strengthening the connections between sign-based representations and written language.

In a recent meta-analysis, Suggate (2016) suggested that long-term effects of reading interventions are actually quite restricted, with a small average effect size of $d = .22$ across different intervention types (Cohen's d is a standardized effect size and can be regarded as small above .20, medium sized above .50, and large when moving beyond .80; Cohen, 1992). Nevertheless, a positive effect indicates that at least some pupils benefit from interventions, which may be important at a practical level. However, when comparing different types of interventions targeting phoneme-letter correspondence, phonology-word correspondence, fluency, or reading comprehension strategies, the strongest effects were found for reading comprehension strategy interventions, and the other groups did actually not seem to have any long-term effects (Suggate, 2016). Based on Suggate's (2016) definitions, Omega-is is a reading comprehension intervention. In

summary, then, Omega-is-d2 may be particular likely to produce positive effects on reading development in DHH signing children.

Summary and Preliminary Models

Several aspects of sign language and cognition are investigated in the present work, and are put in relation to developing reading skills. The literature suggests that computerized sign language based literacy training may support developing reading skills in DHH signing children. Proposed associations between sign-based representations and access to meaning when DHH signing children are learning to read are depicted in Figure 3. Strong connections exist between meaning and both phonological and orthographic forms (filled arrows). However, for DHH signing children who are still learning to read, there may be no connections at all or only weak connections between phonological and orthographic forms (unfilled arrow), making access to the meaning of written language unreliable. These connections are likely to be supported by Omega-is-d2 training.

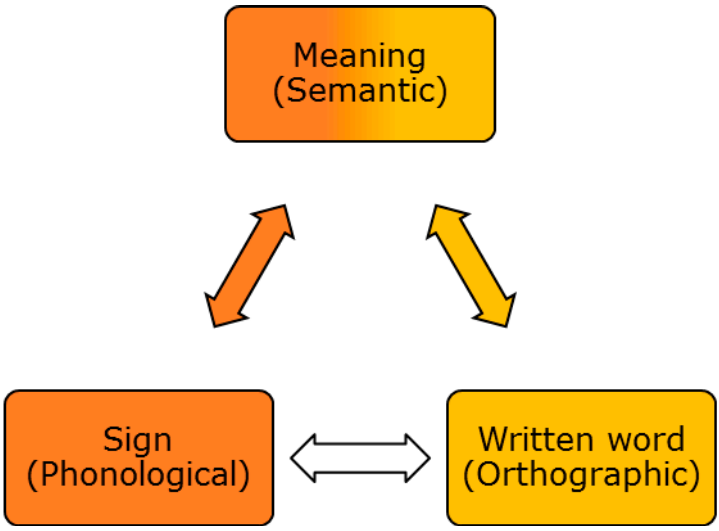

Figure 3. Theoretical model of the relationship between meaning (semantic representations), sign language (phonological representations), and written words (potential orthographic representations). The figure is inspired by a similar model by Hermans, Knoors, Ormel, and Verhoeven (2008a).

Further, sign language PA may be associated with developing word reading skills and sign language comprehension with developing reading comprehension. Working memory capacity is likely to be related to developing reading skills both at word and text level, and ToM may be related to reading comprehension. Finally, imitation of unfamiliar signs may reveal mechanisms of importance for developing word reading in sign language users, and imitation of familiar signs – as a proxy for processing of semantic representations (i.e., vocabulary) – is likely to be related to reading comprehension development. The associations of particular interest in the present work are depicted schematically in Figure 4 (word reading) and Figure 5 (reading comprehension).

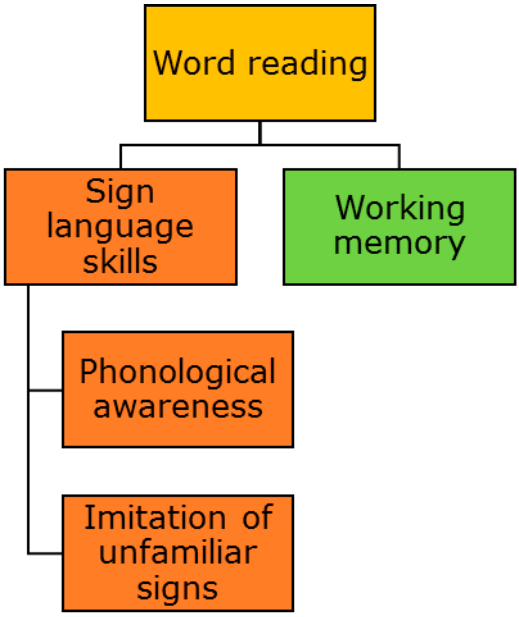

Figure 4. A preliminary model of the associations between word reading and sign language (phonological awareness and imitation of unfamiliar signs) and cognitive (working memory) skills based on theoretical considerations and empirical observations.

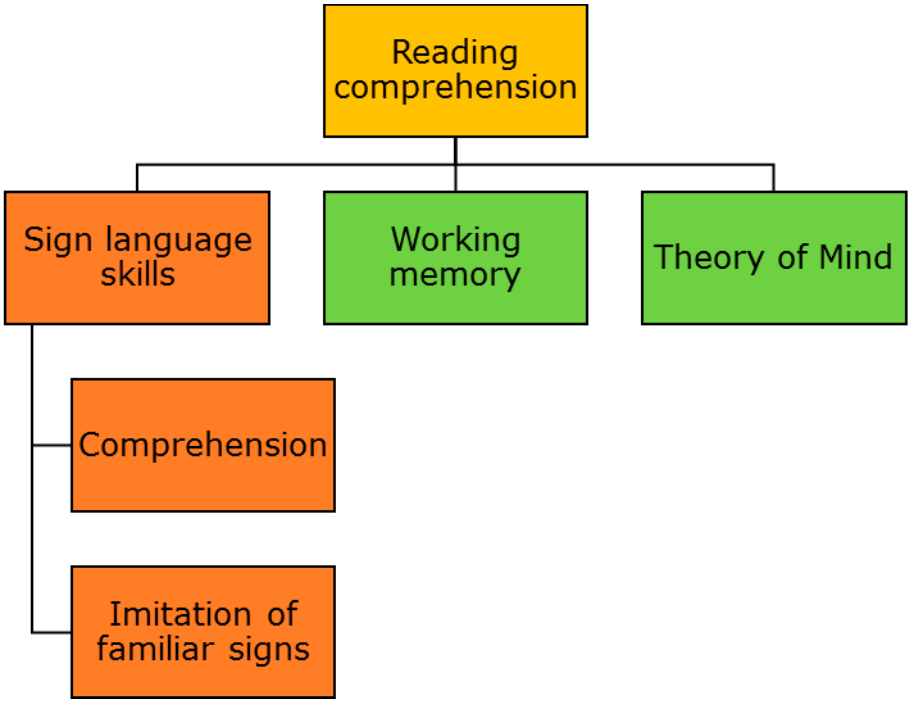

Figure 5. A preliminary model of the associations between reading comprehension and sign language (comprehension and imitation of familiar signs) and cognitive (working memory and Theory of Mind) skills based on theoretical considerations and empirical observations.

Aims and predictions

In this work, the primary aim was to determine whether word reading and reading comprehension can be improved in DHH signing children who are learning to read by training the link between sign and written language. Another aim was to investigate concurrent and longitudinal associations between sign language, cognitive skills and reading skills in this population. It was predicted that:

- The Omega-is-d2 intervention would lead to improved reading skills;
- Sign language PA and imitation of unfamiliar signs would be positively associated with word reading;
- ToM, working memory, sign language comprehension and imitation of familiar signs (i.e., vocabulary) were all predicted to be positively related to reading comprehension.

Summary of Empirical Studies

This thesis includes four empirical studies that are reported in four articles:

- In Paper I, the aim was to investigate whether spoken language PA or sign language PA were related to word reading in children who are learning to read and who are pupils at RSNS. It was predicted that sign language PA would be positively associated with word reading. No specific prediction was made for spoken language PA.
- The aims of Paper II were to explore how sign language skills influence the precision of imitating manual gestures that vary in phonological and semantic content, and how precision of imitation relates to language and cognitive skills in signing and hearing non-signing children. It was predicted that 1) sign language experience would reveal more precise imitation of manual gestures than for sign naïve children at first presentation, and that 2) sign naïve children would shrink the gap to signing children at the second presentation, due to establishment of new representations. Further, 3) imitation of familiar signs would be performed with higher precision than that of unfamiliar signs, and 4) both groups would imitate lexical manual gestures with higher precision than non-lexical manual gestures. Finally, 5) language skills were predicted to be positively correlated with imitative precision in both groups.
- In Paper III, the aims were to investigate ToM development in RSNS pupils, as well as the interrelations between ToM, working memory, sign language comprehension, and reading comprehension in this group. Reading comprehension was expected to be predicted by ToM, working memory and sign language comprehension.
- Finally, in Paper IV the aims were to evaluate the effects of Omega-is-d2 training on word reading and reading comprehension, and to determine how sign language skills at phonological, semantic, and comprehension levels related to developing reading skills in RSNS pupils who are learning to read.

Methods

Participants

Two groups were included in the present project: children who attend RSNS and who are learning to read (Papers I-IV), and Grade 1 hearing non-signing children (Paper I and II). The latter group acted as a comparison group and word reading skills (assessed with a standardized task, *Wordchains*, Jacobson, 2001) did not differ between the two groups. All five RSNS in Sweden were invited to participate in the present project, and two accepted this invitation. The criterion for admission to these schools is HL (The National Agency for Special Needs Education and Schools, 2016). During the period in which the data collection for the work described in this thesis was carried out (2012-2013) the total population at these five schools was 352 individuals (Swedish National Agency for Education, 2015). DHH participants were sampled from this context to ensure that all used SSL. Hearing participants were sampled from four different schools in a municipality in southeast Sweden with representative socioeconomic status. Raw scores on selected reading measures across participants are presented in Table 1.

Table 1: Age and Group Average Performances on Wordchains (WC) and Woodock Passage Reading Comprehension (WPRC) for Deaf and Hard-of-Hearing (DHH) Participants Without and With Additional Disabilities (DHH, AD), and for Hearing Participants

		Age		WC		WPRC		Papers
	N	*M*	*SD*	*M*	*SD*	*M*	*SD*	
DHH	13	10	2.3	7.2	4.8	3.8	1.2	I, II, III, IV
DHH, AD	3	10	1.8	2.3	4.0	1.7	0.6	IV
Hearing	36	7.5	0.3	8.3	4.3	14	8.8	I, II

Participants and their parents provided informed consent, which was attested in writing by the parents, and the project was approved by the Regional Ethical Review Board in Linköping, Sweden (dnr 2012/192-31). Background data was collected from the parents of the participants by questionnaire (for both DHH and hearing participants) or interview (only for DHH participants). In some cases the parents omitted to provide information and thus background data is incomplete.

Deaf and hard-of-hearing participants

Staff members at participating schools identified 17 potential participants that were at an early stage of reading development; that is, they did not yet read fluently, but showed an interest in text and were able to identify written words at a level corresponding to typical readers in Grade 1. This was a critical aspect, since Omega-is typically is used with children who are still learning to read, and not with children who can already read. One potential participants was excluded because of no HL despite attendance at one of the participating schools. Three further potential participants had an additional medical or developmental disability (see Table 1), and their data were only included in one of the papers (Paper IV).

Deaf and hard-of-hearing participants without additional disabilities

The main group for the present project consisted of 13 DHH signing children (7 girls and 6 boys) from grades 1-7 with a mean age of 10.2 years (*SD* = 2.3). The wide age range reflects the variability in the degree of difficulty DHH pupils experience in learning to read (Lederberg et al., 2013; Mayberry, del Giudice, & Lieberman, 2011; Trezek et al., 2011). Eleven used technical aids and thus had at least some access to speech: five used only HA (four bilateral); five used only CI (four bilateral) and one had a CI on one ear and a HA on the other. Based on ten reports, the mean age of fitting of technical aids was 4.1 years (*SD* = 2.3). Up-to-date audiological records were not available and because sign language skills, cognitive skills and reading development were at the focus of the project, audiological measurements were not made. Two of the participants had a vision deficit which was corrected.

Nine participants primarily used SSL for communication, and four of them had at least one deaf native signing parent. Based on the six available parental reports for these nine individuals, mean age of first exposure to SSL was 2.8 years (*SD* = 3.3, range 0.0-8.0), and mean age of first exposure to Swedish was 2.4 years (*SD* = 3.3, range 0.0-8.0). Three participants used both SSL and Swedish. Data for these three individuals showed that the mean age of first exposure to SSL was 4.3 years (*SD* = 1.8, range 3.0-6.3), and the mean age of first exposure to Swedish was 2.0 years (*SD* = 3.5, range 0.0-6.0). Finally, one participant used SSL and another spoken language (age of first exposure to SSL and Swedish was 11.7 years). All participants used

SSL in school. Seven of the participants were born abroad, one in an expatriate family; age at which residence in Sweden commenced ranged from 2.2 to 10.6 years, based on five available parental reports. None of the participants born elsewhere originated from the same country. The primary languages spoken in the participants' homes were SSL (n = 4), a mixture of SSL and Swedish (n = 3) or a spoken language from Central Asia (n = 1), Central Europe (n = 1), and the Middle East (n = 1). Raven's Coloured Progressive Matrices (RCPM) (Raven & Raven, 1994) was used to screen for non-verbal cognitive ability; one scored one point below the 5th percentile of what was expected for that age group, and the rest above. For ten participants, the educational level of the mother was reported: one had less than nine years of primary education, six had three years of secondary education, and three also had post-secondary education, one with a degree.

Hearing participants

For Paper I and II, a group consisting of thirty-six typically developing children (20 girls) with no reported HI or knowledge of sign language attending first grade of primary school were included for comparison purposes. The mean age of the group at the first test occasion was 7.5 years (SD = 0.3). One of the participants had corrected to normal vision, and all had Swedish as their first language. All scored above the 5th percentile on RCPM (Raven & Raven, 1994). The educational level of the mother was less than nine years of primary education for one, three years of secondary education for eleven, and post-secondary education for 23 (19 had a degree).

Methodological considerations

When designing the present project, what tests to include was carefully considered, as well as who would to be assigned as test leader (fluency in SSL was a demand), and how to implement Omega-is-d2 at the participating schools. Given the scarcity of cognitive and language tests developed with DHH signing children in mind, both in Sweden and internationally, test selection was a difficult process. Further, the heterogeneity of this population in Sweden (Svartholm, 2010) made this process even more difficult, since heterogeneity might lead to larger variability in ability than can be captured by a test. On the other hand, test leader selection and implementation of the intervention progressed with relative ease, thanks to enthusiastic staff members at participating schools.

Assessment in deaf and hard-of-hearing signing children

Assessment of psychological constructs is a question of validity. In a general sense, validity refers to whether performance on a specific task or test can be assumed to reflect the individual's level on a specific psychological construct (Cronbach & Meehl, 1955; Strauss & Smith, 2009). More specifically, validity has to do with both the theoretical assumptions underlying the measurement as well as aspects of using a specific measure to assess a specific construct in a specific population (Strauss & Smith, 2009). Importantly, validity does not generalize across populations. Instead validity refers to the meaning of a test score of a specific individual within a specific setting (Messick, 1995). When cognitive or language tests are used across different populations, regarding, e.g., culture, language use, or biological disposition, there is a risk of measurement invariance (van de Vijver & Leung, 2011), which in essence means that the test might not measure the same psychological construct across populations, at least not with the same precision. Most cognitive and language tests are developed for use with hearing individuals. Thus, using the same test with DHH signing children might threaten the validity of the test (c.f., Morere, 2013). It is possible to investigate whether tests are invariant across populations, but such research is resource and time-consuming. For the present work, these issues were handled by motivating test selection based on theory, earlier research, and practical experiences.

Selection of tests and development of test battery

At the start of the present project, no standardized measures of language or cognitive skills for DHH signing children were available in Sweden. Thus, when selecting measures to include, a first step was to carefully survey and consider what measures had been used in earlier research in other countries on similar groups, or are used by practitioners working with this population in Sweden. This led to identification of a set of tests to use, but some new tests also had to be constructed. The full test list, when assessment where carried out with each test and in which paper they are used is displayed in Table 2.

Tests of language

An SSL adaption of the BSL Receptive Skills Test (Herman, Holmes, & Woll, 1999) was developed for a Swedish context by the National Agency

for Special Needs Education and Schools in parallel to data collection in the present project. This test is one of few internationally established tests of sign language comprehension (Enns & Herman, 2011; Haug, 2011; Johnston, 2004), and was used as a measure of this in the present work.

To assess other aspects of SSL skills, relating to phonological and semantic processing, new tests were developed (i.e., a sign language phonological awareness test and an imitation task, described below), since no established tests of such skills were identified prior to data collection. Hearing participants were administered the Phonological processing subtest of NEPSY (Korkman, Kirk, & Kemp, 1998).

Tests of reading

In Sweden, Wordchains (Jacobson, 2001) is an established test of word reading in DHH signing children (Hendar, 2004; Petersson et al., 2000). Further, lexical decision tasks have been used to assess word reading skills with similar groups in earlier research (e.g., Merrills, Underwood, & Wood, 1994; Transler & Reitsma, 2005).

An earlier version of DLS Bas (Järpsten, 2004) was used by Heiling (1994) in a group of deaf children, and similar tests, i.e., in which written text is matched to pictures, are commonly used with this population in Sweden (Petersson et al., 2000), but also internationally (Mayberry, del Giudice, et al., 2011). The Woodcock Passage Reading Comprehension test (Woodcock, 1998) is commonly used as a measure of reading comprehension in DHH groups, both internationally (Mayberry, del Giudice, et al., 2011) and in Sweden (e.g., Nakeva von Mentzer et al., 2014).

Table 2: Overview of Tests Used for Deaf and Hard-of-Hearing (DHH) and Hearing Participants, What They Measure and in Which Papers They Are Used

Test	DMDX	Measures	Paper(s)
Language			
C-PhAT-SSL (see Paper I)	X	*PA of SSL*	I-IV
C-PhAT-Swed (see Paper I)	X	*PA of Swedish*	I, II
Phonological processing (Korkman et al., 1998)		*PA of Swedish*	I
SSL Receptive Skills Test (Herman et al., 1999)		*SSL comprehension*	I-IV
Reading			
Lexical decision (e.g., Transler & Reitsma, 2005)	X	*Word reading*	I, II, IV
Wordchains (Jacobson, 2001)		*Word reading*	I, II, IV
DLS Bas (Järpsten, 2004)		*Reading comprehension*	IV
WPRC (Woodcock, 1998)		*Reading comprehension*	II, III, IV
Cognition			
Imitation task[a] (see Paper II)	X	*Imitation/Sign language skills*	II, IV
RCPM (Raven & Raven, 1994)		*Non-verbal cognitive ability*	I-IV
Simon task[b] (Lu & Proctor, 1995)	X	*Inhibition*	-
Clown test (Birberg Thornberg, 2011)		*Working memory*	I-IV
Theory of Mind scale (Wellman & Liu, 2004)		*Theory of Mind*	III
Control variables			
Bead threading (White et al., 2006)		*Motor control*	II
Button pressing (see Paper I)	X	*Motor speed*	I
Digit and Letter decision (see Paper I)	X	*Print knowledge*	I
Response matching (see Paper I)	X	*Cognitive speed*	I

DMDX = a cross marks that the test was administered in DMDX, a computer based display system widely used for linguistic and cognitive experiments (Forster & Forster, 2003); C-PhAT = Cross-modal Phonological Awareness Test, Swedish Sign Langauge version (C-PhAT-SSL) and Swedish version (C-PhAT-Swed); SSL = Swedish Sign Language; WPRC = Woodcock Passage Reading Comprehension; RCPM = Raven's Coloured Progressive Matrices; PA = phonological awareness.
[a]Can be regarded as a measure of language skills for deaf and hard-of-hearing signing participants.
[b]This task was not included in papers due to unreliable results.

Tests of cognition

RCPM (Raven & Raven, 1994) has been used in earlier research on similar populations (e.g., Jones, Gutierrez, & Ludlow, 2015; Rudner et al., 2015; Woolfe, Want, & Siegal, 2002), and is commonly used in practice with this population in Sweden. The RCPM is typically regarded as a valid screener of non-verbal cognitive ability (Nisbett et al., 2012).

To assess working memory capacity, a visuo-spatial task called the Clown test (Birberg Thornberg, 2011; Sundqvist & Rönnberg, 2010) was used. Birberg Thornberg (2011) developed this task based on the Mr. Peanut Man task (Kemps, de Rammelaere, & Desmet, 2000). A visuo-spatial task, without any explicit utterances to remember, was used in favor of a sign-based task of working memory to reduce the risk of confounding factors relating to individual differences in language ability.

In addition to a working memory task, a task tapping onto executive skills (i.e., inhibition; described in Rudner et al., 2015) was also included in the design (see Table 2). Due to unreliable results on this task it was not included in any of the papers.

ToM was assessed with a SSL version of a ToM scale (Wellman & Liu, 2004) that has been used in several studies of DHH children (Peterson et al., 2005; 2012; 2016; Remmel & Peters, 2009). The SSL version is based on a Swedish version of the scale (Sundqvist, Koch, et al., 2014), and was adapted for SSL with support from both an experienced sign language interpreter and staff members at collaborating schools (see Paper III for more details).

Bead threading (White et al., 2006) was one of several control tasks, and had earlier been used by Marshall, Denmark and Morgan (2006) in a similar group. A similar task is also included in one of the few cognitive test batteries developed to be used within a deaf population, the *Hiskey-Nebraska Test of Learning Aptitude* (Hiskey, 1966).

Development of computerized test battery

In the present work, PA of sign language was one of the targeted sign language skills. However, no such task was available, at least not for children who use SSL, and thus we developed an experimental task to assess this: the Cross-modal Phonological Awareness Test (C-PhAT; briefly described below and in detail in Paper I). Further, we also wanted to explore how the precision of imitation of manual gestures was influenced by prior

representations and related to cognitive and language skills, and thus an experimental imitation task was designed (described in detail in Paper II). In addition, a number of control tasks were constructed to ensure that participants understood the general test procedure of (i.e., Button pressing, Response matching) and were familiar with the stimuli in C-PhAT (i.e., Digit and Letter decision). These tasks also allowed the participants to become familiarized with the test procedure of the C-PhAT.

All computerized tasks were created for use in presentation software DMDX (version 4.1.2.0; Forster & Forster, 2003). DMDX is widely used platform for linguistic and cognitive experiments and registers timing and responses with high accuracy (Garaizar, Vadillo, López-de-Ipiña, & Matute, 2014). Unless otherwise stated, stimuli were presented as black text in capital letters of 115 points in Times New Roman, on a white background. Participants responded by pressing one white and one black Jelly Bean Twist button (6.5 cm in diameter), that always corresponded to the same responses ("yes" and "no"). The "yes" button was placed to the side of the participant's dominant hand.

Control tasks

To make sure that the testing procedure of the C-PhAT was suitable for participants, four control tasks were developed (described more in detail in Paper I): a Button pressing task, a Response matching task, and Digit and Letter decision tasks. In the Button pressing task, participants pressed a designated button thirty times as fast as they could. This was mainly for familiarization with response buttons, but mean button pressing time in s was also used as a control measure of motor speed in Paper I. The second control task, a Stimuli matching task, helped the participant to learn what button corresponded to a yes and no responses respectively. The mean response time in *s* on this task was also used as a control measure in Paper I. The Digit and Letter decision tasks were used in Paper I to screen for participants' ability to identify correctly oriented digits and letters.

Cross-modal Phonological Awareness Test (C-PhAT)

A key theoretical notion behind the C-PhAT is that phonology may be regarded as the sub-lexical structure of any form of language (Andrews & Wang, 2015; Brentari, 2011). This notion is supported by a growing amount of empirical work (e.g., Andin et al., 2014; Berent et al., 2014;

Brentari, 2011; Corina, Hafer, et al., 2014; Gutiérrez, Müller, et al., 2012; Gutiérrez, Williams, et al., 2012; MacSweeney, Waters, et al., 2008; McQuarrie & Abbott, 2013). In a Swedish context, Andin et al., (2014) created a task in which the labels of stimuli could share handshapes in the Swedish manual alphabet or manual numeral systems or rimes in speech. Andin et al. (2014) argued that this task relied on sign language PA. Others have utilized similar tasks to measure sign language PA in a North American (Corina, Hafer, et al., 2014; McQuarrie & Abbott, 2013) or British (MacSweeney, Waters, et al., 2008) context. The C-PhAT is an extension of work by Andin et al. (2014), and can be used to assess both sign language PA and spoken language PA.

In C-PhAT, pairs of printed letters or a printed letter and digit are presented on a computer screen (see Table 3). When administered as a measure of sign language PA (C-PhAT-SSL), the task is to decide for each pair whether or not the printed letters and digits share handshape. When administered as a measure of spoken language PA (C-PhAT-Swed), the task is to decide for each pair whether or not the spoken phonological labels for the printed letters and digits rhyme. Both accuracy, calculated as *d'* which corrects for guessing (Swets, Tanner, & Birdsall, 1961), and mean response time in s for correct responses can be used as dependent measures on the task. For the present project *d'* was used, since a number of participants had near chance performance, which would make response time a less sensitive measure, that is, if a participant's responses are random and fast, they would confound results.

Table 3: Examples of Stimuli Pairs (1, 2, and 3) in the Cross-modal Phonological Awareness Test and Their Phonological Labels in Swedish and the Swedish Manual Alphabet and Manual Numeral Systems (SMS)

	1		2		3	
Stimuli (print)	6	X	P	H	J	7
Swedish	/sɛks/	/ɛks/	/peː/	/hoː/	/jiː/	/ɧʉː/
SMS						

1 = labels rhyme in Swedish; 2 = handshapes are shared in the Swedish manual systems; 3 = no similarity in handshapes in the Swedish manual systems, or in Swedish labels.

Imitation of manual gestures

To investigate how imitation precision of manual gestures is influenced by sign language skills, and how imitation of different types of manual gestures relate to developing reading skills, a new imitation task was developed for the present work. A set consisting of three familiar signs, three unfamiliar signs, and three non-signs, was selected from an available data base of video recorded manual gestures (see Cardin et al., 2016; Orfanidou, Adam, McQueen, & Morgan, 2009; Rudner, Orfanidou, Cardin, Capek, Woll, & Rönnberg, 2016). Familiar signs were real signs in SSL, thus, invoking both phonological and semantic representations for DHH participants (i.e., vocabulary). Unfamiliar signs were signs from BSL which were phonologically legal but had no meaning in SSL (comparable to unfamiliar words for hearing children, Marshall, 2014). The last category, i.e., non-signs, bore reduced phonological information and violated some phonological characteristic of both SSL and BSL (non-lexical gestures). In the imitation task, the nine videos are presented in random order on a computer screen, and after each video the participant is instructed "Now, it is your turn". This is often used as a prompt for imitation (e.g., Wang, Williamson, & Meltzoff, 2015). Test sessions were video recorded, and the correspondence between the manual gestures presented in the video clips and the participants' responses were rated on a later occasion.

The Omega-is-d2: A new sign language version of Omega-is

A pilot of a SSL version of Omega-is, the Omega-is-d1, was created in a master's thesis project (Hermansson, 2011). In the project, words and sentences from the program was translated into SSL and video recordings of the translations were incorporated into Omega-is. The results from the master thesis was later published (Rudner et al., 2015). However, based on the results in Rudner et al.'s (2015) study, it was not possible to determine whether or not Omega-is-d1 had a positive effect on reading skills. There were some limitations that might have contributed to this. In particular, participants only worked with the program for two weeks, that is, for a maximum of ten days. In comparison to its predecessor, the Omega-is-d1 was also incomplete. In particular, due to technical issues, animations had to be dropped, and only one, two and three word sentences were translated into SSL, covering only around 15% of the total material from the original

program. For the present work, SSL videos of all words and sentences were recorded, and both videos and animations were included in a new sign language version of Omega-is: the Omega-is-d2. The SSL video material was created in collaboration with the Sign Language Section of the Department of Linguistics, Stockholm University. Further, the training period was extended from two to four weeks. These changes were assumed to increase the likelihood of a positive effect on reading development. How the Omega-is-d2 works, and how it was constructed is further described in Paper IV.

Study design

For RSNS pupils, a cross-over intervention design was combined with a longitudinal design. When assessment was carried out in relation to the intervention is displayed in Figure 6 (also, see Table 2, for list of tests, and Table 4, for an overview of which tests were used when for DHH respectively hearing participants). An initial test occasion (T1) was followed by a baseline period, which in turn was followed by a cross-over intervention period that began with a second test occasion (T2) and ended with the fourth test occasion (T4). The cross-over period involved two separate training periods, in which the participants used the Omega-is-d2 (see Figure 6). DHH participants were placed into two groups, one that first used Omega-is-d2 (T2-T3) and then attended regular schoolwork (T3-T4), and one that first attended regular schoolwork (T2-T3) and then worked with Omega-is-d2 (T3-T4). Time intervals were unequal due to practical reasons (described in Paper IV), but, importantly, the amount of training that participants received did not differ across groups. At the point where the groups switched conditions, a third test occasion was placed (T3). Finally, a fifth and last test occasion (T5) was placed nine months after the beginning of the study, and approximately six months after the end of the cross-over period. Hearing participants, on the other hand, were only part of the longitudinal design. That is, they were assessed on the first (T1) and last (T5) test occasion and did not receive Omega-is-d2 training. This combined design made it possible to investigate concurrent and longitudinal relations between reading skills and predictor variables, between group (i.e., DHH and hearing participants) performance on study variables, as well as effects of Omega-is-d2 training on developing reading skills for RSNS pupils.

	Longitudinal design			
	Description	**Occasion**	**Time**	**Assessment**
		T1	Week 0	Language, cognition and reading
	Baseline period			
		T2	Week 5	Reading
Cros-over design	*Cross-over period 1* Group 1: Omega-is-d2 training Group 2: Schoolwork			
		T3	Week 10	Reading
	Cross-over period 2 Group 1: Schoolwork Group 2: Omega-is-d2 training			
		T4	Week 16	Reading
	Follow-up period			
		T5	Week 39	Language, cognition and reading

Figure 6. The combined longitudinal and cross-over study design for deaf and hard-of-hearing participants.

Procedure

All participants were tested individually in a quiet room at their schools. For DHH participants, test administrators were recruited from participating schools, to ensure fluency in SSL, experience from working with DHH signing children and familiarity with participants. To ensure that all tests were administered similarly across the administrators, written instructions were available both in SSL and in Swedish. SSL instructions were based on a formalized system for coding Swedish into SSL (Bergman, 2012). There were three test administrators who administered all tests except the test of SSL comprehension. The test of SSL comprehension was administered by two individuals who were specially trained to administer this test.

Four undergraduate students and the author did the testing of the hearing participants, and the same written instructions in Swedish that were available for test administrators working with DHH participants were used. Before data collection began, the author trained the undergraduate students in the testing procedure, by providing instructions and demonstrating testing, but also feedback on pilot sessions. For both samples there was a recommended test order. However, pauses and changes to test order were allowed when needed to optimize participant's performances. At which test occasions tests were administered is displayed in Table 4.

Table 4: An Overview of Test Occasions at Which the Tests Were Administered to Deaf and Hard-of-Hearing (DHH) and to Hearing Participants

Test	DHH					Hearing	
		Cross-over period					
	T1	T2	T3	T4	T5	T1	T5
Language skills							
C-PhAT-SSL	X				X		
C-PhAT-Swed	X				X	X	X
Phonological processing							X
SSL Receptive Skills Test	X				X		
Reading skills							
Wordchains	X	X	X	X	X	X	X
Lexical decision	X	X	X	X	X	X	X
DLS Bas	X	X	X	X	X		
Woodock Passage Reading Comprehension	X	X	X	X	X	X	X
Cognition							
Imitation task	X				X	X	X
Raven's Coloured Progressive Matrices	X				X	X	
The Simon task	X				X		
The Clown test	X				X	X	
Theory of Mind scale	X				X		
Control variables							
Bead threading	X				X	X	X
Button pressing	X				X	X	X
Digit and Letter decision	X				X	X	X
Response matching	X				X	X	

C-PhAT-SSL = Cross-modal Phonological Awareness Test, Swedish Sign Language version. C-PhAT-Swed = Cross-modal Phonological Awareness Test, Swedish version. SSL = Swedish Sign Language.

Omega-is-d2 training

Omega-is-d2 training was implemented as a part of DHH participant's daily schoolwork, and participants were instructed to use Omega-is-d2 for 10 minutes per school-day for four weeks (i.e., 20 days), which was the total time of a training period. Before the cross-over period commenced, two matched groups were created from the sample and the order in which groups received Omega-is-d2 training was randomized. Teachers were encouraged to sit and work together with participants or at least be available to them and assist them when needed. However, due to practical reasons, this was not a requirement.

Statistical considerations

Throughout this work, no corrections were made for multiple statistical tests. The main reason for this was that the small sample size would basically lead to no power to detect any significant relationships between variables, or differences between groups, regardless of the size of the effects. Thus, correcting for multiple tests would lead to a great risk for Type II errors. At the same time, a small sample size leads to an increased risk of random effects or strong influence of outliers. These risks were handled by grounding our tests on prior theory and empirical observations, statistically exploring violations to normality and comparing results from non-parametric and parametric methods, as well as investigating scatterplots for correlations. Finally, it was always sought to maintain as much data as possible in data analysis.

Results

Paper I

Correlational analysis was performed to explore evidence of validity for the C-PhAT as a measure of PA, and whether spoken language PA or sign language PA predicted word reading in RSNS pupils who are learning to read. Results from hearing participants indicated that the C-PhAT is a valid measure of PA. Performance on C-PhAT-Swed predicted, $r(36) = .54$, $p = .001$, scores on the standardized measure of PA, the Phonological processing subtest from NEPSY (Korkman et al., 1998). Further, performance also was positively correlated with scores on Wordchains, $r(36) = .36$, $p = .030$, and lexical decision, $r(36) = .37$, $p = .028$. Importantly, no associations were observed with basic cognitive variables. DHH signing participants on the other hand, performed at chance level on C-PhAT-Swed, and scores were related to Working memory, $r(13) = .60$, $p = .032$, and Cognitive speed, $r(12) = -.68$, $p = .016$, but not to measures of Word reading. However, for C-PhAT-SSL the pattern was reversed, with strong associations to Wordchains, $r(13) = .66$, $p = .013$, and lexical decision, $r(13) = .63$, $p = .021$, but no association with cognitive performance.

Paper II

A repeated measures split-plot ANOVA, with two within group factors: test occasion (one and two), and type of manual gesture (familiar signs, unfamiliar signs, and non-signs); and one between group factor (DHH signing and hearing non-signing participants), was conducted to investigate the effects of sign language experience on imitation of manual gestures. Further, correlational analysis between imitative precision and language and cognitive skills was performed. For the ANOVA, all main effects were statistically significant, as well as the interaction between test occasion and group. Exploration of simple main effects revealed that both DHH and hearing participants imitated the manual gestures with higher precision on the second than on the first presentation. Further, the between group difference was not statistically significant at the first presentation, but it was at the second. Thus, DHH signing participants had no initial advantage over the hearing non-signing participants on the task, despite the difference in experience of sign language across groups, but DHH participants did have a steeper development on the task over test occasions. The correlational analysis indicated that language skills predicted performance on the imitation task once representations had become established, and that both language processes at the phonological, semantic and comprehension levels were involved in imitation of manual gestures in DHH participants, but only at a comprehension level for hearing participants. Cognitive performance did not predict imitative precision in neither group.

Paper III

Performance of RSNS pupils on Wellman and Liu's (Wellman & Liu, 2004) ToM scale was compared between participants with parents who primarily used SSL (n = 4) and those with parents who did not (n = 9). It was also compared to performance of DHH signing children from an Australian context reported in earlier studies (Peterson et al., 2005, 2012). Correlational analysis was also performed between ToM scale performance, sign language comprehension, working memory capacity and reading comprehension. In spite of stronger sign language skills among DHH participants with parents who primarily used SSL, no difference on ToM was detected from DHH participants with parents who used a spoken language. Further, overall group performance was worse than for native signing DHH children of similar age reported in earlier studies from an Australian context (e.g., Peterson et al.,

2005), but comparable to late signing DHH children and younger typically developing hearing children from the same studies. Positive correlations were revealed between ToM, working memory, and reading comprehension. The association between ToM and reading comprehension was still statistically significant after controlling for general language skills, $r_p(10) = .63$, $p = .028$.

Paper IV

Two hierarchical linear regression analyses (Singer & Willett, 2003) were conducted to investigate whether word reading and reading comprehension improved between the first and the last test occasion for the RSNS pupils, to evaluate the effect of Omega-is-d2 training on reading development, as well as to investigate whether sign language skills predicted individual growth in reading skills over time. It was revealed that both word reading and reading comprehension improved over time. Further, results suggested that Omega-is-d2 had a positive effect on word reading but not on reading comprehension. Individual growth in word reading appeared to be predicted by precise imitation of unfamiliar signs. Further, there was also a trending association between development in reading comprehension and precise imitation of familiar signs (i.e., vocabulary).

General Discussion

In the present work, the effects of sign language based computerized literacy training on developing reading skills were evaluated in children who are learning to read and who are RSNS pupils. Further, concurrent and longitudinal associations between reading skills and sign language and cognitive skills (i.e., working memory, imitation, and Theory of Mind) were investigated. Whether sign language skill is associated with imitation precision of manual gestures and ToM ability was also investigated. The main prediction was that sign language based computerized literacy training would produce positive effects on reading skills. Further, sign language PA and imitation of unfamiliar signs were predicted to be associated with word reading. ToM, working memory, sign language comprehension, and imitation of familiar signs (i.e., vocabulary) were all predicted to be positively related to reading comprehension. Results provided partial support for the predictions. Sign-based literacy training appeared to have a positive effect on word reading. Also, specific sign language skills were concurrently and longitudinally related to developing reading skills, and reading comprehension was concurrently associated with both ToM and working memory.

Developing Reading in Deaf and Hard-of-Hearing Signing Children

In the present work, evidence of the involvement of sign language skills in the early stages of reading for DHH signing children was most convincing at the level of word reading. However, some results also indicated a positive connection between sign language skills and reading comprehension. In Paper I, sign language PA was associated with word reading. The associations between imitation of manual gestures and reading skills were investigated more in detail in Paper IV. Results in Paper IV indicated that imitation of familiar signs predicted the developmental trajectory in reading comprehension, that imitation of unfamiliar signs predicted development of word reading, and a possible effect of Omega-is-d2 training on word reading. However, contrary to prediction, no training effect was observed on reading comprehension (Paper IV), and no convincing association was found between sign language comprehension and reading comprehension (Paper III, IV).

Sign language and word reading

Results from the present work indicate a connection between word reading and sign language PA (Paper I) and that sign-based representations can become associated with orthographic forms (Paper IV). However, spoken language PA was not associated with word reading (Paper I). Instead, the ability to make correct speech-based rhyme decisions the present study was strongly associated with working memory and cognitive speed, suggesting that tasks that invoke speech-based representations involve basic cognitive rather than specialized language based processing in DHH signing children (c.f., McQuarrie & Parrila, 2009). However, others have indicated that speech based representations is associated with reading in DHH signing children (e.g., Kyle et al., 2015).

Earlier research on word reading in DHH signing children indicates an important role for sign language skills relating to sub-lexical and lexical processing. In particular, sign language PA has been reported to be correlated with word reading (McQuarrie & Abbott, 2013), and experimental evidence indicates that signs can become automatically associated with their orthographic counterparts (e.g., Morford et al., 2011; Ormel et al., 2012; Pan et al., 2015). In addition, fingerspelling ability, that is, proficiency in accessing representations of and producing the handshapes that correspond to written letters in a manual alphabet, may facilitate establishment of new written vocabulary (Haptonstall-Nykaza & Schick, 2007), and has been found to be related to reading skills (Stone et al., 2015). Further, exercising connections between signs and written words seems to facilitate word reading (Reitsma, 2009; Wauters et al., 2001). Thus, both earlier work (e.g., Ormel et al., 2012) and the present findings lend support to theoretical notions suggesting that mapping between sign-based representations and written words (Crume, 2013; Haptonstall-Nykaza & Schick, 2007; Hermans et al., 2008a; Hoffmeister & Caldwell-Harris, 2014) may be an important part of early word reading development in DHH signing children. However, the present results may help us to understand these connections a little better.

Sign language PA was concurrently but not longitudinally related to word reading (Paper I and IV), and identical results were revealed for working memory (Paper IV). However, longitudinally, word reading was associated with the imitation of unfamiliar signs, which taps into both sign-based representations and working memory capacity (Marshall, 2014). Earlier studies on hearing children indicate that performance on analogous speech based tasks, that is, imitation of unfamiliar words, is linked to word

learning (Gathercole, 2006) and reading ability (Melby-Lervåg & Lervåg, 2012; Pennington & Bishop, 2014). Further, an association between the precision of repeating unfamiliar words and the ability to read words has also been reported in children with CIs who primarily use speech (Dillon & Pisoni, 2006; Nakeva von Mentzer et al., 2015). The ability to retrieve stored sub-lexical representations and assemble these into a new lexical item in working memory, as reflected by an ability to precisely imitate unfamiliar lexical forms (c.f., Marshall, 2014), might reflect a propensity for change in the lexical system, or lexical restructuring (Metsala, 1999). The association between imitation of unfamiliar signs and word reading development in the present work suggests that this process may be a particularly important mechanism for developing word reading in DHH signing children; perhaps more important than for hearing children, since DHH children who primarily use sign language establish new representations as they learn to read. Based on the present results, it is not possible to determine what form such new representations take in long-term memory for DHH signing children, e.g., sign-based or orthographic. However, the present findings do indicate that the mechanism involved is amodal in nature, given that the association here is between sign-based and speech-based language. Although intriguing, the notion that this mechanism is amodal in nature is a speculative claim that needs to be tested in future studies.

One important aspect to consider is that the effect of Omega-is-d2 training on word reading in Paper IV was evaluated on written language material that had not been specifically practiced. This indicates that training effects transfer to general word reading skills. It might be that sign-based representations are used to support identification of written words. Yet another possibility is that the ability to process orthographic forms has developed. The latter idea is in line with theoretical notions suggesting that DHH signing children learn to read by cracking the orthographic system (e.g., Bélanger & Rayner, 2015).

Sign language and reading comprehension

Word reading and reading comprehension have been identified as connected, but at the same time distinct, processes (Hulme & Snowling, 2014; Kamhi & Catts, 2012) and were regarded as such in the present work. The results of the Omega-is-d2 intervention (Paper IV) and patterns of associations (Papers I-IV) indicate that this distinction generalizes to DHH signing children. Relative to Grade 1 hearing children, RSNS pupils performed better on word

reading than on reading comprehension (see Methods section). A similar pattern has been reported by others (e.g., Wauters, Bon, & Tellings, 2006). Further, development in word reading was steeper than in reading comprehension. The divide between word reading and reading comprehension is sometimes underemphasized in the literature on DHH children (Andrews & Wang, 2015). The findings in the present work indicate that it is important to separate these levels of reading in the case of DHH signing children who are learning to read.

The interface between lexical items and their meaning has been suggested to play a key role in reading comprehension (Perfetti & Stafura, 2014). It is well established that vocabulary size is related to reading development in DHH children (Lederberg et al., 2013), and some empirical observations also indicate an association in DHH signing children (e.g., Hermans et al., 2008b). In line with these earlier observations, there was a trending association between the precision of imitation of familiar signs, which might be regarded as a proxy for semantic representations (i.e., vocabulary), and development in reading comprehension in Paper IV. Thus, vocabulary may be a bottle neck for developing reading comprehension in DHH signing children. This notion fits well with the remark that DHH children, who in general represent a group that displays difficulties in learning to comprehend texts (Lederberg et al., 2013), tend to have weaker vocabulary development than hearing children (Lederberg et al., 2013; Lund, 2016). This suggests that the critical issue when DHH signing children learn to read might not be a lack of the right type of phonological representations, but rather the right amount of semantic representations. Learning to read words is a good start, but comprehension cannot be achieved without the appropriate semantic representations by which to understand them.

Surprisingly, no statistically significant association between sign language comprehension and reading comprehension was observed in Paper III. On the other hand, when a different method was used for analyzing this association in Paper IV (i.e., combining the two measures of reading comprehension into an index and analyzing the data with hierarchical linear modeling), results indicated a possible association at the beginning of the study (i.e., at week 0). Sign language comprehension did not, however, predict development in reading comprehension over the 39 weeks between the first and final test occasions. The results regarding a connection between sign language comprehension and reading comprehension was thus somewhat mixed in the present work. Prior studies have reported associations

between sign language comprehension and reading comprehension (e.g., Chamberlain & Mayberry, 2008; Freel et al., 2011), and in a meta-analysis by Mayberry, del Giudice, et al. (2011) general language skill was the strongest predictor of reading ability in DHH children. Further, even though languages differ in their perceptual demands and rely on representations in different modalities, comprehension processes are likely to rely on similar cognitive mechanisms across all languages and language modalities (Cardin et al., 2013; MacSweeney, Capek, et al., 2008; Siegal, 2004). Thus, it may be that some factor relating to the heterogeneity of the sample or choice of language comprehension measures can explain why no associations could be established in Paper III. In Paper III, both reading comprehension and sign language comprehension measures showed trending extreme end value effects, i.e., reading comprehension scores were close to floor level, and sign language comprehension was close to ceiling level. With such restrictions in variability, the likelihood of detecting associations decreases. Another potential issue is that sampling beginning readers regardless of grade, might lead to special sample characteristics. As noted by Hoffmeister and Caldwell-Harris (2014), DHH signing children learn to comprehend speech-based language as they learn to read. The DHH participants in the present work did not yet comprehend written language but they did comprehend sign language, and it may thus be incorrect to interpret the association as one between two comparable measures of language comprehension.

Taken together, these results indicate that sign language comprehension does not automatically transfer into reading comprehension (Paper III, IV), but that specific sign language skills (i.e., vocabulary) may support development of reading comprehension (Paper IV). This overall pattern is in line with notions on restricted automatic transfer of sign language skills to reading skills (Goldin-Meadow & Mayberry, 2001; Holzinger & Fellinger, 2014), and recent findings indicating a key role of semantic processing in reading comprehension in deaf individuals (Hirshorn et al., 2015). Further, it also aligns with studies of cross-modal second language development, indicating that prior representations in one modality support the establishment of language representations in another modality (e.g., Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012).

Cognition and Language in Deaf and Hard-of-Hearing Signing Children

Besides investigating the intersection between sign language and reading skills, the present work also investigated how sign language skill relates to imitation precision of manual gestures (Paper II) and ToM ability (Paper III). Furthermore, associations between working memory, ToM and both sign-based and speech-based language skills were investigated.

Sign language and imitation of manual gestures

The ELU model (Rönnberg et al., 2013) predicts that language processing is more efficient when the incoming signal can be matched exactly to a prior representation (familiar lexical forms), than when there is only a partial (unfamiliar lexical forms) or minimal (non-lexical forms) match. Similar predictions can be made based on associative theories of imitation (Heyes, 2016). Hearing children typically repeat familiar lexical forms more precisely than unfamiliar lexical forms (e.g., Dispaldro et al., 2011), and it appears that it is easier for them to form representations of unfamiliar but legal lexical forms than speech strings that violate the phonology of their native language (e.g., Morra & Camba, 2009). Further, correct imitation of unfamiliar lexical forms in the manual modality was reported to be more likely for deaf signing children than for hearing non-signing children by Mann et al. (2010). The results in the present work (Paper II) do not, however, fully align with these notions and earlier findings. In particular, no evidence of more precise imitation of manual gestures for signing than for non-signing children was observed at the first presentation (T1) of the specific set of gestures used in the present work. Further, signing participants did not imitate familiar signs with higher precision than unfamiliar signs. Thus, no evidence of an initial advantage of sign language experience was found, or of a separation between familiar and unfamiliar lexical forms. However, sign language skill did seem to provide a basis for stronger establishment of object specific representations, since signing children improved more than non-signing children between the first and second test occasion. This was also suggested by the correlational pattern, which indicated that associations between sign language skills and imitation was more convincing longitudinally than concurrently. In addition, language comprehension predicted change in imitation precision for hearing participants over time, suggesting than some process relating to language comprehension predicts the establishment of

new manual representations for sign naïve children. Associations between language skills and gestural imitation have also been reported in earlier studies, at both a behavioral (e.g., Farrant, Maybery, & Fletcher, 2011) and a neural (e.g., Kühn, Brass, & Gallinat, 2013) level, and has been suggested to reflect shared reliance on representation of sequential information (Kühn et al., 2013). This connection should be further investigated in future work.

Imitation precision of familiar and unfamiliar signs

The phonology of a sign language often carries semantic information (Thompson, Vinson, Woll, & Vigliocco, 2012), and some earlier studies indicate that semantic information, in addition to phonological, does not influence the processing efficiency of sign-based stimuli in deaf adults (e.g., Cardin et al., 2016; Rudner et al., 2016). This may explain why no difference in performance was found between familiar and unfamiliar signs for DHH signing children in the present study. In contrast to spoken language users (Marslen-Wilson, 1987), it may be the case that for sign language users, semantic representation does not provide any further constraints on the lexical target beyond the influence of phonology. Since semantic and phonological representations seem to independently support language processing in hearing adults (Rönnberg et al., 2013) and children (e.g., Dispaldro et al., 2011), earlier findings (e.g., Cardin et al., 2016) and the present findings suggest that the relationship between phonology and semantics might differ across sign language and spoken language. This may in turn have implications for language processing and development, but also cognitive development. Studying cognitive development across children with and without HL who use sign language or speech or both, may help us further understand how the interface between perception and production of a language influence both language development and cognitive development.

Gesture type and a surprising effect of sign language experience

Recent findings indicate that non-lexical manual gestures are more difficult to process than are lexical manual gestures both for signers and non-signers (Cardin et al., 2016; Rudner et al., 2016). These earlier findings relating to adults were here replicated for children. Thus, it seems that it is more demanding to process manual gestures than to break the phonological patterns of signed languages, even for individuals with no previous

knowledge of sign language. As discussed above, this may indicate that the phonological characteristics of a language arise as a consequence of more efficient neural processing of its perception and production (c.f., Cardin et al., 2016).

At the first test occasion, the DHH signing children in the present work were no more precise in their imitation of manual gestures than the hearing non-signing children were. This somewhat surprising finding may indicate that from a certain age children with typical motor functioning might have the motor repertoire needed to produce lexical items from sign language. Although an associative account of imitation suggests that motor expertise, like being a sign language user, should provide an advantage regarding imitation of manual gestures, the theory also suggests that representations that are close enough to a target behavior might be sufficient (Heyes, 2016). On the other hand, sign language experience does seem to lead to changes in the neural processing of any type of manual gestures in adults (Newman et al., 2015; but see, e.g., Cardin et al., 2016). This suggests that with development, a difference between signing and non-signing individuals might emerge. It may be that lexical manual gestures align with motor-repertoires intrinsic in most children, and thus are optimally suited to be processed by a developing cognitive system. Meaningful acts (e.g., mimes of object use) seem to be easier to imitate precisely than novel, meaningless acts (Tessari & Rumiati, 2004). Thus, more precise imitation of lexical manual gestures than non-lexical manual gestures may be caused by differences in the perceived meaningfulness and inherent motor patterns.

Imitation and the Developmental Ease of Language Understanding model

Although the findings relating to imitation of manual gestures in the present work indicate some qualitative differences across signing and sign-naïve children, they do not fully align with the predictions based on the ELU model (Rönnberg et al., 2013). Thus, the findings prompt an adjustment of the ELU model. In Paper II, a Developmental ELU model, the D-ELU, is proposed to take into account the present set of results.

Both the ELU model (Rönnberg et al., 2013) and the D-ELU model propose that when the incoming language signal does not correspond to a prior representation in long-term memory, a qualitative change in processing occurs. More specifically, the system then relies partly on other mechanisms to understand the meaning of the incoming signal. In particular, this process

is constrained by working memory capacity. The ELU model predicts that a mismatch condition invokes domain general semantic representations to aid understanding (i.e., meaning prediction system, Rönnberg et al., 2013). In addition to this, the D-ELU model predicts that domain specific representations (e.g., sign-based representation) are invoked, which in turn provides an opportunity either for redefining already established representations or establishing completely new meaning-based representations (c.f. Kuhl, 1991). Although this process may be invoked without the occurrence of an imitative act, imitation may increase the likelihood for lexical restructuring by strengthening the association between perception and production (c.f., Heyes, 2016). The D-ELU model may provide a base for more developmentally focused research within CHS.

Theory of Mind, language, and working memory

It is well established that language skill and ToM are developmentally connected in hearing (Carlson et al., 2013; Milligan et al., 2007), DHH signing children (Lederberg et al., 2013; Peterson, 2009), and DHH children with CI who primarily use spoken language (Sundqvist & Heimann, 2014), and a connection between ToM and working memory has also been reported in the literature (Davis & Pratt, 1995; Gordon & Olson, 1998; Meristo & Hjelmquist, 2009; Mutter, Alcorn, & Welsh, 2006). Further, working memory capacity is typically involved in language comprehension (Kintsch & Rawson, 2007), particularly when understanding is hard to achieve (Rönnberg et al., 2013). In Paper III, a positive association between working memory and ToM was observed, in line with theoretical ideas suggesting that working memory supports successful ToM (e.g., Siegal & Varley, 2002), and earlier findings (e.g., Meristo & Hjelmquist, 2009). However, sign language comprehension was not related to working memory or ToM. Given the strong support for connections between these variables in the literature, the present results were likely due to restricted power, some aspect pertaining to heterogeneity of the sample or the methods used. The trending ceiling effect on the sign language comprehension task suggests that this tasks does not reflect a condition under which comprehension was challenging for the RSNS pupils in the present work, and this might explain why no connection to working memory was found (c.f., Rönnberg et al., 2013). The results of the present work further suggest that children attending RSNS have typical progression in their development of ToM according to the Wellman and Liu (2004) scale, albeit delayed in this particular sample. Similar findings have

been reported for DHH signing children in other cultural settings where English is the ambient spoken language (e.g., Peterson et al., 2005) but this is the first time that such results have been generalized to a setting where another spoken language predominates.

A connection between Theory of Mind and reading comprehension

This is the first work in which a positive association between ToM and reading comprehension is reported in DHH signing children who are learning to read. However, earlier studies have reported associations between ToM and reading skills in hearing children (e.g., Astington & Pelletier, 2005; Kim, 2015a; Miller et al., 2013; Ricketts et al., 2013) and adults (Mar, 2011). Such overlaps have been interpreted in several different ways, for example, reflecting the involvement of general language skill (e.g., Astington & Pelletier, 2005) or working memory capacity (e.g., Miller et al., 2013) in both ToM and reading. Other studies have reported that there might be a unique association between ToM and reading comprehension after controlling for both general language skills (e.g., Ricketts et al., 2013) and working memory (e.g., Kim, 2015a). Thus, the connection between ToM and reading comprehension observed in the present work might involve more than an overlap with general language skills or working memory capacity. It is tentatively suggested that the pattern of associations may reflect the ability to draw appropriate inferences for constructing an adequate representational model in working memory.

Inference making is a key mechanism in both ToM and reading comprehension (Kim, 2015a; Ricketts et al., 2013). Deaf children have been shown to be better at literal reading comprehension than inferential reading comprehension, and inference making actually also seems to be challenging for many of these children in sign language (for a review, see Marschark & Wauters, 2008). Thus, drawing appropriate inferences may be challenging under in any language for at least some DHH signing children, and any process that builds on making inferences based on certain knowledge that is not perceptually accessible (e.g., verbal or gestural behavior) at a given time might thus be difficult to solve (c.f. Courtin et al., 2008). Specifically for reading comprehension, there are some promising findings suggesting that reading interventions involving strategies for making text inferences may increase reading comprehension in DHH signing children (e.g., van Staden,

2013; Walker et al., 1998). Focused inference making training might support both text and mind reading in this population.

Working memory and developing reading skills

According to the ELU model (Rönnberg et al., 2013), under challenging language understanding conditions, processing becomes constrained by working memory capacity. Further, flexible resource models of working memory suggest that when it is more difficult to form representations, it may be harder to process them in working memory (Ma et al., 2014). Thus, working memory is likely to become involved when children are learning to read. This applies in particular for DHH signing children who are learning to read in a second language in a modality other than that of their preferred language. Prior studies have reported positive associations between working memory capacity and reading skills in both hearing (National Institute for Literacy, 2008) and DHH signing children (e.g., Daza et al., 2014). In the present study, some support for concurrent associations between working memory and both word reading and reading comprehension was found (Paper III, IV). This suggests that working memory may be involved in identification of written words and support construction of a representational model of written text while DHH signing children are reading, but it does not seem to support development of these skills.

Even though working memory capacity is involved in any mental activity that unfolds over time (Diamond, 2013), it does not always indicate separation between individuals. For example, if all individuals in a particular group have developed beyond a critical threshold of working memory capacity that is needed to solve a specific task, more of the same may not support further development of the skills underlying task performance. Such skills may include the ability to manipulate specific types of representations of varying quality in working memory. The findings relating to word reading and reading comprehension discussed above, indicate such developmental specialization. Further, in relation to reading, it might be of importance to distinguish between processes that support the development of reading skills, and processes that support reading activities. Working memory might always be involved in constructing a representational model of an incoming language signal (Kintsch & Rawson, 2007; Zwaan, 2015). However, mechanisms of learning are more likely to depend on specific processes tapping into the interactions between working memory capacity and long-term memory systems (c.f., Gathercole, 2006).

Self-regulation and academic skills

It has been suggested that self-regulation, involving social skills, like ToM, as well as working memory and executive skills, may provide a basis for learning all kinds of academic skills, and that some adequate level of self-regulatory skills is needed to satisfy the necessary conditions for learning (Blair & Raver, 2015; Ziv, 2013). On the other hand, learning conditions for children with weak self-regulatory skills might be enhanced if environmental adjustments were made (Blair & Raver, 2015). The empirical support for such broad models includes studies indicating that individuals with weak self-regulation typically perform poorly in school, in particular in reading development (Trzesniewski, Moffitt, Caspi, Taylor, & Maughan, 2006). Further support comes from studies reporting positive effects of interventions targeting language specific, cognitive and social skills on several academic skills, including reading (Bierman, Nix, Greenberg, Blair, & Domitrovich, 2008). Thus, in the case of DHH signing children, it may be important to focus not only on the skills in which the "deficits" become apparent, like reading, but try to take a broader perspective on skewed development (c.f., Andrews & Wang, 2015; Nelson, 1998).

Limitations

The sample of DHH signing children in the present work was small and heterogeneous. A small sample size is always difficult to handle in statistical analysis, especially when the individuals within the sample vary on several characteristics that constitute nuisance variables. More specifically, it is unlikely that statistical estimates correspond to population parameters, and error terms are likely to be biased. Thus, results from the present work should be interpreted with caution and need to be replicated in future studies. Further, the size of the sample constrained statistical power. In relation to the aims of this study, this restricted the possibility to detect effects of Omega-is-d2 training and the possibility to disentangle the relative contribution of each variable in relation to reading development. Although heterogeneous and small, the characteristics of DHH participants in the present work were similar to descriptions of RSNS pupils in the literature (Svartholm, 2010). Further, of the total population at the participating schools, approximately 5% participated, and since only pupils who were at an early stage of reading development were targeted, the sample is likely to be a major part of the total

population available at the time of the study. This suggests that the sample is likely to be representative of the targeted population.

Another potential issue is that the number of test leaders (N = 3 for all tests apart from the test of SSL comprehension, N = 2) was large in relation to the number of RSNS pupils (N = 16). It is likely that different test leaders administer tests differently, and this may influence performances. On the other hand, test leaders were selected on the basis that they should be fluent in SSL and familiar with participants. It was assumed that this would lead to optimal conditions for participants to perform, and a standardized set of test instructions was used to keep instructions similar between test leaders.

When interventions are provided in parallel to regular reading instruction, effects are known to be weaker than when interventions are provided in addition to regular instructions (Suggate, 2016). The implementation of Omega-is-d2 was thus not optimized in the present study, since it was integrated as a part of DHH participants' regular schooling. Further, compliance to the procedure was not strictly monitored, although log files from Omega-is-d2 provided information on how participants had worked with the program. Further, the speech material which is included as standard in the regular version of Omega-is, was deleted from the Omega-is-d2 in the present work to avoid the possible confounding effects of access to speech material during training for participants who used some speech. Based on the theoretical framework behind Omega-is (Nelson, 1998), input in more modalities scaffolds learning and increases the time information is handled by working memory, and thus also the likelihood for establishing new representations. In an educational setting, it is recommended to apply all modalities that may provide the specific pupil with an aid of understanding.

In the present work, most analyses involve correlational data, and besides the effect of Omega-is-d2 training, the results relating to word reading is no exception to this. As indicated above, estimation of correlation coefficients is unreliable in small samples, which calls for cautious interpretations of results. Further, correlational data is difficult to interpret in terms of underlying mechanism (e.g., Strauss & Smith, 2009), albeit at the same time may indicate where to look for causal mechanisms. In this work a CHS (Arlinger et al., 2009) framework was applied, and the particular focus was on mechanisms at a psychological level. Even though CHS provides a useful meta-theoretical framework for studies relating to populations with HL, at the same time it also constrains interpretation of behavioral data in terms of

underlying psychological mechanisms (c.f., Slife & Williamson, 1995). This mainly represents a limitation at a theoretical level.

Future Research

Interactions between biological (e.g., hearing thresholds), psychological (e.g., cognitive and language skills), and social (e.g., home environment, schooling) factors are often acknowledged in the literature on psychological development in DHH children (e.g., Lederberg et al., 2013). This approach can be further utilized in relation to language and cognitive development in DHH signing children in future work. For example, our knowledge of what factors beyond the biological level that predict cognitive and language development in DHH children is sparse (Campbell et al., 2014). On a psychological level, one important avenue for future research is to learn more about the way in which perceptual and cognitive processes during infancy predict later language outcomes in DHH children. This applies for DHH children who primarily use speech and for children who primarily rely on sign language. There are several cognitive paradigms that can be used with infants during the first year of life (e.g., Baillargeon et al., 2016; Bauer, 2006; Rovee-Collier & Giles, 2010), and can thus be used even before implantation of CI (Kral & Sharma, 2012). Of particular interest in relation to the present work are imitation paradigms, which have been used to assess both social (e.g., Oostenbroek, Slaughter, Nielsen, & Suddendorf, 2013) and cognitive (e.g., Sundqvist, Nordqvist, Koch, & Heimann, 2016) skills in infants. Imitation paradigms could be utilized to investigate how short- and long-term memory systems interact with language input in the manual-visual and oral-aural modality as language develops in DHH infants. Such behavioral methods could be used in combination with eye-tracking (e.g., Óturai, Kolling, & Knopf, 2013), and/or electroencephalography (e.g., Nordqvist, Rudner, Johansson, Lindgren, & Heimann, 2015), to help us isolate key steps in language processing and mechanisms of language development in different modalities.

Imitation of different types of lexical forms, e.g., familiar or unfamiliar, is often used as measures of phonological and/or semantic processing for hearing children (e.g., Sundström et al., 2014). However, in the case of DHH children, these type of tests has hitherto been studied only to a limited extent (e.g., Dillon & Pisoni, 2006; Meier, 1987). In future work, investigating imitation of utterances across hearing and DHH individuals, both signing and

non-signing, may reveal how language specific, in particular, phonological, and domain general, e.g., semantic, representations, and processes related to manipulation of these (e.g., working memory) support reading development.

The D-ELU model has to be further tested in future work. In particular, defining what constitutes a redefinition of established respectively establishing a completely new representation is important, since these two processes may rely on different mechanisms. This might initially be a theoretical hurdle, but has to be operationalized for empirical testing for further development of the model. In addition, whether the actual repetition of a behavioral act (i.e., imitation) has an additional value in establishing/redefining representations beyond that of inner imaginary of the act could be tested experimentally. Perhaps, actually repeating the behavioral act may be particularly important for establishing representations in populations where this may be difficult to achieve, for example, in children with HL who only can perceive a degraded spoken language signal. As indicated in earlier (e.g., Cardin et al., 2016) and the present work, the relationship between phonological and semantic representations may differ across modalities, and this may influence language development. Investigating behavioral (e.g., imitation) changes and their neurobiological substrates in developing language systems cross-modally, e.g., in deaf infants who receive CIs and learn both sign language and speech, or adults who know speech and who are learning sign language (e.g., sign language interpreters), may answer some questions relating to this issue. Another interesting area is how the proposed learning mechanism in the D-ELU model, that is, manipulation of the lexical system invoked by imitation of lexical forms, is fostered within social experience.

Future research should also focus on interventions to support reading development in DHH signing children. In particular, it is important to move beyond single words and to focus more on comprehension processes. Eye-tracking and electroencephalography may be particularly valuable in this process, given that these methods effectively used to detect disruptions in the reading process due to phonological, orthographic, or semantic interference effects (Bélanger & Rayner, 2015; Leinenger, 2014). One specific aspect to investigate further in relation to DHH signing children's reading comprehension is inference-making ability. In paper III it is suggested that the link between ToM and reading comprehension may be due to an underlying mechanism based on inference-making ability. This idea should be investigated in future studies. In particular, whether problems with

inference-making in DHH signing children (Marschark & Wauters, 2008) reflect a domain general mechanism involved in ToM, sign language comprehension and reading comprehension, or if it reveal itself in a specific domain due to lack of relevant background knowledge (e.g., mental state vocabulary for ToM) should be investigated. Inference-making may also be an appropriate focus for future reading comprehension interventions (c.f., Walker et al., 1998), and could be tested in combination with Omega-is-d2 training.

Further development and evaluation of the C-PhAT, both as a measure of spoken language PA and of sign language PA, as well as the imitation task is warranted. Future studies should investigate whether DHH signing children with better reading skills than those in the present sample, perform better at the C-PhAT-SSL and imitate manual gestures with better precision. If phonological processing of sign language is related to developing word reading skills, as suggested here, DHH signing children who are better word readers than the participants in the present work should perform better at the C-PhAT-SSL and imitate unfamiliar signs with better precision. For DHH signing children who perform better on reading comprehension but not word reading tasks, it is hypothesized that vocabulary skills, as indicated by more precise imitation of familiar signs, will be stronger. This would be in line with the notion that semantic representations are particularly important for developing reading comprehension. The C-PhAT may be suitable for a tactile modality and thus possible to use with Braille readers. By doing so, PA can be assessed using the same test material in groups that read in the visual and groups that read in the tactile modality (e.g., blind children). This would lead to a new way of testing the idea that PA reflect an amodal mechanism. Further, the C-PhAT-Swed was recently tested as a measure of PA in proficient readers by using response time rather than *d'* as the dependent variable. Unpublished data from our lab suggest that using the test in this way provides valid estimates of spoken language PA in hearing adults. For example, performance on the C-PhAT-Swed seems to differ between native speakers of Swedish and adults who are learning Swedish as their second language. However, the practical utility of the C-PhAT is to a great extent determined by how well it can detect children at risk of atypical language or reading development. Given the simplicity of the task, it might be useful for children younger than those included in the present work.

The effectiveness of Omega-is-d2 should also be further investigated in future studies. In the present study, effects of training were tested on

standardized and experimental reading tests (i.e., transfer effects). Future studies should in addition to this include structured testing of the word-sign connections worked with in the program, to evaluate the effects of Omega-is-d2 on establishing cross-modal connections (c.f., Wauters et al., 2001). Future studies could also manipulate the amount and intensity of training, which would likely lead to a better understanding of how Omega-is-d2 is optimally implemented in an educational setting.

Key Findings, Implications and New Models

In this work, the primary aim was to determine whether word reading and reading comprehension can be improved in DHH signing children who are learning to read by training the link between sign and written language. Another aim was to investigate concurrent and longitudinal associations between sign language, cognitive skills and reading skills in this population. The findings revealed that:

- In line with the initial prediction, computerized sign language based literacy training appeared to have a positive effect on developing word reading. Omega-is-d2 may be a useful tool in an educational setting.
- As predicted, sign language PA and imitation of unfamiliar signs were related to word reading, and imitation of familiar signs (i.e., vocabulary) was positively related to reading comprehension. Phonological processing may reflect an amodal domain of language skills of particular importance for word identification and lexical restructuring.
- Also in line with the predictions, ToM, working memory, and imitation of familiar signs (i.e., vocabulary) were all positively related to reading comprehension, and a marginally significant association between imitation of familiar signs and developing reading comprehension was found. Semantic processing may be a key aspect of developing reading comprehension in DHH signing children, and inference making constrained by working memory capacity, may explain the association between comprehension of minds and texts in this group. Interventions aimed at establishing a rich vocabulary and learning to apply prior knowledge in different situations may be useful for supporting reading development and ToM.

- A qualitative difference in the imitation of manual gestures between signing and sign naïve children was indicated by the results. A modified version of the ELU model (Rönnberg et al., 2013), the D-ELU model, is proposed to account for the pattern of findings.
- Typical, although delayed, progression in ToM was observed. The role of inference making in ToM development in DHH signing children should be investigated in future studies.

The results from the present project line up with empirical observations (e.g., Hermans et al., 2008b; Wauters et al., 2001) and theoretical notions (e.g., Goldin-Meadow & Mayberry, 2001; Hoffmeister & Caldwell-Harris, 2014) suggesting that sign-based representations do support reading development in DHH signing children. Thus, representations, and activities related to their manipulation and use, aid processing of language, even when the surface form of the language is based on another modality. This is a key notion in the ELU model (Rönnberg et al., 2013), and also the D-ELU model (Paper II). However, the D-ELU model provides a developmental focus that makes it more useful for understanding language development.

The preliminary model (Figure 3) discussed in the introduction is adapted in the light of the results of the intervention in the present work. It schematically depicts the connections between sign-based, orthographic and semantic representations (see Figure 7). In spite of a possible effect on word reading (indicated by the filled arrow between phonological and orthographic representations), meaning (semantic representations) of written language was still generally inaccessible via written language (as indicated by the unfilled arrow between orthographic and semantic representations). Thus, the form of written language may be accurately recognized, but understanding the content of that surface form is restricted.

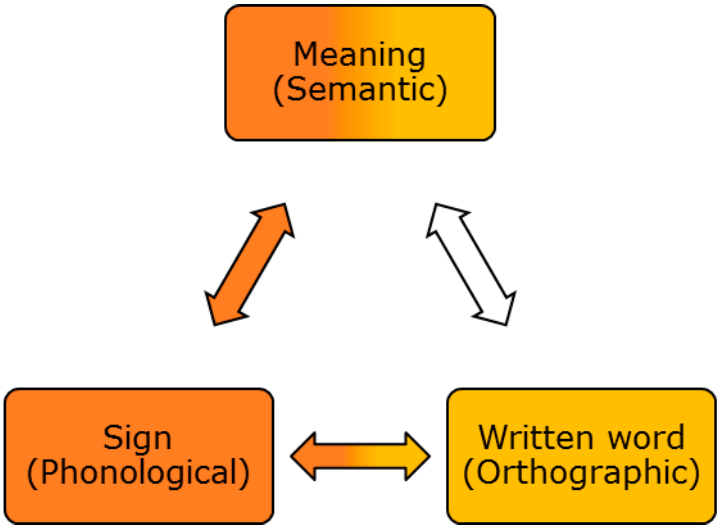

Figure 7. A schematic model of the effects of Omega-is-d2 training.

A model of the associations between sign language skills, cognition and reading skills is presented in Figure 8. This model is a revised version of the models presented in the introduction (Figure 4 and Figure 5), and is based on the present set of results put in relation to the broader literature. Working memory (WM) can be regarded as a capacity limit of a cognitive system that is involved in any type of processing that occurs over short time intervals. This is indicated by the one-way arrows from WM to reading comprehension, word reading, and ToM (Theory of Mind). WM is a layer of the cognitive system that connects old and new experiences. This is signified by the arrows

indicating a route from stored representations (phonological, Pho, and semantic, Sem, representations) to word reading and reading comprehension via WM. Word reading involves the matching of orthographic forms to stored representations. A match reflects a connection that may be perceptually unimodal (orthographic form-orthographic representation) or more abstract, i.e., cross-modal, and is the starting point of comprehension. The activation and manipulation of stored representations in WM that occur when a language signal is analyzed may also lead to redefinition of the lexical system or establishment of completely new lexical items. Routes involved in such developmental effects are represented by double lined arrows. The bidirectional arrow between reading comprehension and ToM indicates that the nature of this association is undefined.

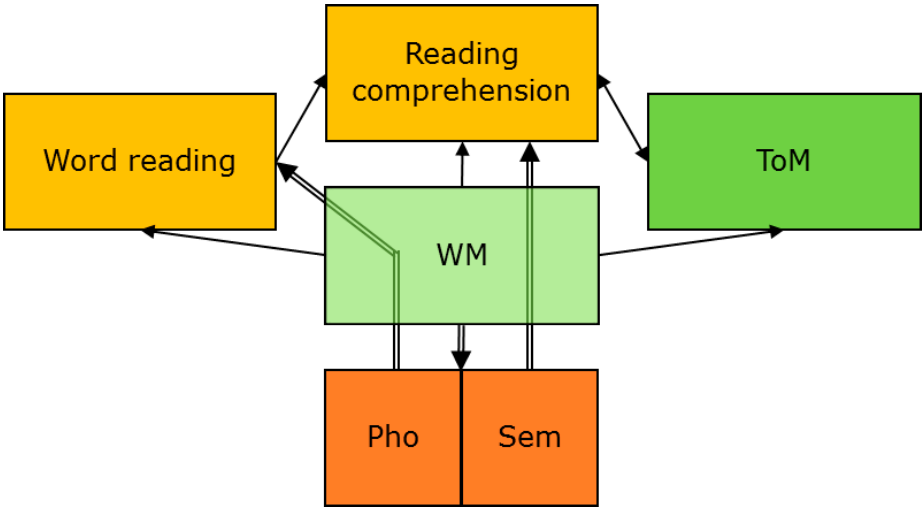

Figure 8. Literacy, Developmental Ease of Language Understanding, and Theory of Mind (ToM).

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