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Computerized Sign Language-Based Literacy Training for Deaf and Hard-of-Hearing Children

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Abstract

Strengthening the connections between sign language and written language may improve reading skills in deaf and hard-of-hearing (DHH) signing children. The main aim of the present study was to investigate whether computerized sign language-based literacy training improves reading skills in DHH signing children who are learning to read. Further, longitudinal associations between sign language skills and developing reading skills were investigated. Participants were recruited from Swedish state special schools for DHH children, where pupils are taught in both sign language and spoken language. Reading skills were assessed at five occasions and the intervention was implemented in a cross-over design. Results indicated that reading skills improved over time and that development of word reading was predicted by the ability to imitate unfamiliar lexical signs, but there was only weak evidence that it was supported by the intervention. These results demonstrate for the first time a longitudinal link between sign-based abilities and word reading in DHH signing children who are learning to read. We suggest that the active construction of novel lexical forms may be a supramodal mechanism underlying word reading development.

Proficiency in sign language may provide a foundation for learning to read in deaf and hard-of-hearing (DHH) children who use sign language as their primary mode of communication (Chamberlain & Mayberry, 2000; Goldin-Meadow & Mayberry, 2001; Hoffmeister & Caldwell-Harris, 2014). For example, it has been suggested that DHH signing children learn the meaning of orthographic forms by connecting them to sign-based representations (Crume, 2013; Hermans, Knoors, Ormel, & Verhoeven, 2008a; Hoffmeister & Caldwell-Harris, 2014). Indeed, both experimental (Ormel, Hermans, Knoors, & Vervhoeven, 2012) and correlational (Hermans, Knoors, Ormel, & Verhoeven, 2008b) data indicate a connection between sign language and reading skills in DHH signing children who are learning to read. However, this idea has seldom been utilized as a basis for reading interventions (for reviews on interventions, see Luckner & Handley, 2008; Luckner, Sebald, Cooney, Young, & Muir, 2005; Tucci, Trussell, & Easterbrooks, 2014). Further, published training studies suggest that training might support improved reading of targeted words (Reitsma, 2009; Wauters, Knoors, Vervloed, & Aarnoutse, 2001), but it is not known whether training effects also generalize to the broader domains of word reading and reading comprehension. The main aim of the present study was to determine whether strengthening the link between the written language and sign language using a computerized literacy intervention improves word reading and reading comprehension in DHH signing children who are learning to read. Related to this, a second aim was to investigate longitudinal associations between sign language skills and reading development in this group.

The intervention in the present study is based on Omega-interactive sentences (Omega-is; Heimann, Lundälv, Tjus, & Nelson, 2004), which is a top-down, or comprehension focused, form of reading intervention (Suggate, 2016). The program has

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several predecessors (Heimann et al., 2004), and is based on Rare Event Transactional Theory (Nelson, 1998; Nelson, Welsh, Camarata, Tjus, & Heimann, 2001). A key notion in this theory is that learning is influenced by several contextual factors, and that a learning situation where all factors converge optimally, is a rare event (Nelson, 1998). For example, motivational materials promote sustained attention to the object of learning, thus increasing the probability that learning will occur. In addition, if such materials relate to relevant prior representations the probability of learning is further increased (Nelson, 1998). In Omega-is (Nelson et al., 2001; Tjus & Heimann, 2000), nouns and propositions are presented as single written and spoken words and sentences with corresponding animations.

Multi-modal presentation of the same content within a short time period is designed to support activation of and attention to relevant representations (Nelson, Heimann, & Tjus, 1997). This is explicitly assumed to ease working memory processing demands, since cognitive resources that would otherwise have been used for semantic retrieval can be used for language processing, and devoted to comparing written language and meaning instead. To promote interest for a wide range of learners, the sentences are of varying length and may include, for example, adjectives, conjunctions, and prepositions, as well as nouns and verbs. Further, the program includes both plausible (e.g., The girl dances) and implausible events (e.g., The lion feeds the penguin), which is assumed to make it more fun and stimulating to work with (Nelson et al., 2001).

Training in Omega-is is comprehension focused, but the conditions for learning are assumed to be equal across all domains of reading (Nelson, 1998). This means that comprehension focused interventions which are sufficiently motivational may improve several reading (e.g., word reading) or reading-related (e.g., phonological awareness) skills and that any such effects may generalize beyond the specific materials used in the intervention. Indeed, in a recent meta-analysis, Suggate (2016) reported that comprehension focused literacy training might actually lead to long-lasting effects not only on reading comprehension but also on, for example, phonological awareness (PA), word reading, and spelling.

Omega-is and its predecessors have been shown to support reading development in children with reading difficulties or with diagnoses associated with delayed reading. These groups include poor readers (Fälth, Gustafson, Tjus, Heimann, & Svensson, 2013; Gustafson, Fälth, Svensson, Tjus, & Heimann, 2011; Helland, Tjus, Hovden, Ofte, & Heimann, 2011), children with autism spectrum disorders, cerebral palsy and hearing loss (Heimann, Nelson, Tjus, & Gillberg, 1995; Tjus, Heimann, & Nelson, 1998, 2004), and profoundly deaf children (Prinz & Nelson, 1985). In particular, Omega-is seems to be effective for children who are struggling at an early reading level. For example, Gustafson et al. (2011) showed that hearing children with reading disability in Grade 2 improved both their word reading and reading comprehension after only twenty-five 15–25 min sessions of Omega-is training. In addition, predecessors to Omega-is, involving a restricted range of exercises (i.e., only up to three word sentences), have shown positive effects on reading development in children with mixed disabilities with reading skills at a preschool level after about 20 sessions of training (Heimann et al., 1995). Thus, it is likely that a sign language version of Omega-is including a wide range of written language material and animations can be used to support reading development in DHH signing children who are just beginning to learn to read.

An earlier study from our lab (Rudner et al., 2015), showed that DHH signing children working with a sign-based, rather than a speech-based, version of this program, the Omega-is-d1, improved their word reading skills. However, the growth in word reading could not be specifically attributed to the intervention (Rudner et al., 2015). We believe that our inability to detect a statistically significant intervention effect was due in part to the relative simplicity of the sentence materials and the lack of animations in Omega-is-d1, and in part to the limited amount of training (10 days) that the participants received.

For the present work, we developed a completely new sign language version of Omega-is: the Omega-is-d2. Like its predecessor, Omega-is-d1 (Rudner et al., 2015), Omega-is-d2 is sign-based but compared to Omega-is-d1 it included more materials with a wider range of complexity and, crucially, it included animations. The animations in Omega-is are designed to support the establishment of connections between the language material and relevant long-term representations. Prior studies suggest that pictorial material aids the establishment of word reading (Reitsma, 2009) and supports reading comprehension (Gentry, Chinn, & Moulton, 2004/2005) in deaf children. Thus, it is likely that a sign language version of Omega-is including a wide range of written language material and animations can be used to support reading development in DHH signing children who are just beginning to learn to read.

Sign languages are natural languages with sub-lexical, lexical, and syntactic structures (Emmorey, 2002). However, sign languages differ from speech-based languages in that they are produced and perceived in the manual-visual channel instead of the oral-aural channel. In spoken languages, the sub-lexical structure relates to place and manner of vocal articulation, whereas in sign languages it relates to the shape, orientation, location, and movements of the signing hands (Brentari, 2011).

In an earlier study, we reported that PA of sign language was concurrently related to word reading in DHH signing children (Holmer, Heimann, & Rudner, 2016a; also, see McQuarrie & Abbott, 2013). PA is typically defined as sensitivity to sub-lexical structure (Wagner & Torgesen, 1987) and this definition applies equally well to spoken and signed language. However, PA for sign language involves sensitivity to contrasts of different articulatory features across signs, such as handshape (Andin, Rönnberg, & Rudner, 2014) or location (MacSweeney, Waters, Brammer, Woll, & Goswami, 2008), whereas PA for spoken language involves, for example, identification of specific phonemes in a word or comparing the sub-lexical structure across different words (Melby-Lervåg, Lyster, & Hulme, 2012). PA for spoken language typically predicts word reading in hearing children (Melby-Lervåg et al., 2012; National Institute for Literacy, 2008). In view of our earlier study (Holmer et al., 2016a), we noted that PA may support the process of connecting prior lexical and sub-lexical representations, regardless of form (i.e., manual-visual or oral-aural), to orthographic forms. Thus, when it comes to development of word reading, sign language PA may reflect the same fundamental ability for DHH signing children that spoken language PA does for hearing children.

Written language is based on spoken language and, thus, there is high correspondence between orthographic forms and speech-based representations (Kamhi & Catts, 2012). This means that hearing children learn to map established spoken language representations to written language when they learn to read (Ziegler & Goswami, 2005). However, there is little micro mapping between the surface representation of a signed proposition and its written equivalent, and syntactically, sign order typically does not follow word order. Thus, DHH signing children have to learn new language structures in order to learn to read (Hoffmeister & Caldwell-Harris, 2014; Svartholm, 2010). In particular, they have to establish new lexical representations, that is, orthographic forms of spoken language (Bélanger &

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Rayner, 2015). This is because all written languages are second languages for these children (Hoffmeister & Caldwell-Harris, 2014; Svartholm, 2010). Little is known about the mechanisms involved in this process for DHH signing children. Studies on hearing children indicate that repetition of unfamiliar words, that is, plausible but meaningless combinations of sub-lexical units, is associated with lexical change (for a review, see Gathercole, 2006), and less accurate repetition is connected to weaknesses in reading skills (Melby-Lervåg & Lervåg, 2012; Pennington & Bishop, 2009). Repetition of unfamiliar lexical forms involves processing and novel arrangement of sub-lexical units in working memory (Gathercole, 2006), and analogous sign-based tasks have been suggested to reflect similar underlying cognitive processes for sign language users (Marshall, 2014). Thus, precise repetition of unfamiliar lexical forms may reflect propensity for lexical restructuring (c.f., Metsala, 1999), and a connection to developing word reading skills may exist in DHH signing children.

Although word reading and, thus, sub-lexical processing skills, are crucial for reading comprehension at early stages of reading (Ripoll Salceda, Alonso, & Castilla-Earls, 2014), language processes beyond the sub-lexical level are likely to come into play during comprehension of written text. These may involve activation of several sources of knowledge (e.g., language specific knowledge and domain general semantic knowledge), as well as appropriate inference making and maintenance of relevant information in working memory (Kamhi & Catts, 2012; Perfetti & Stafura, 2014; Rönnberg et al., 2013). In line with this, Hirshorn, Dye, Hauser, Supalla and Bavelier (2015) recently reported empirical evidence suggesting that maintenance of semantic representations in working memory is a key to reading comprehension in deaf adults. It has also been suggested that general proficiency in sign language is critical for development of reading comprehension in DHH signing children (Chamberlain & Mayberry, 2000; Hoffmeister & Caldwell-Harris, 2014). Stronger sign language skills may enable discussion between pupil and teacher of difficult aspects of reading, which in turn may help the learning child to develop effective reading strategies (Hoffmeister & Caldwell-Harris, 2014). Further, according to the Ease of Language Understanding (ELU) model (Rönnberg et al., 2013), language processing builds on maintenance and updating of a representational model as new information, regardless of modality, enters the system. This processing is constrained by working memory capacity, but also involves domain general semantic knowledge and inference making (also, see Kintsch & Rawson, 2007). Empirical data indicate that general sign language skill is positively related to reading comprehension (Chamberlain & Mayberry, 2008; Schönström, 2010; Stone, Kartheiser, Hauser, Petitto, & Allen, 2015). It may thus be the case that stronger sign language comprehension reflects greater maturity of cognitive mechanisms of relevance for language comprehension in general, which in turn may enable DHH signing children to learn about reading from others.

In a recent study from our lab (Holmer, Heimann, & Rudner, 2016b), participants performed a manual gesture imitation task with three types of gestures: lexical items from Swedish Sign Language (SSL), that is to say, gestures that bore semantic and phonological information (i.e., familiar signs); lexical items from British Sign Language (BSL), that is to say, gestures that bore a large amount phonological information, but had no meaning (i.e., unfamiliar signs); and gestures that had no meaning and bore only a limited amount of phonological information. Even though we refer to the task as an imitation task, in line with Marshall (2014) we regard the task demands as analogous to those involved in repeating familiar and unfamiliar spoken words and non-linguistic utterances. In the present study, the data reported in Holmer et al. (2016b) are reanalyzed to investigate how imitation of unfamiliar signs, that is, representing the processing of sub-lexical units in working memory, and of familiar signs, that is, reflecting processing of semantic representations in working memory, relates to developing reading skills in DHH signing children. Measures of sign language comprehension and sign language PA are also included, as well as a non-linguistic visuo-spatial working memory task that does not involve any explicit language material. The visuo-spatial working memory task was included to access the executive component of working memory (Baddeley, 2012), with no direct influence of language representations. Spoken language skill has been reported to be associated with reading skills in groups of DHH signing children (Niederberger, 2008), and might therefore support reading development. However, we decided against including a measure of spoken language since results from one of our previous studies suggest that the participants in the present study were unable to perform better than chance on a spoken language PA task (Holmer et al., 2016a). When speech representations are weak, they are unlikely to support language development. In the present study, relations between developing reading skills and several different sign language skills and working memory are investigated.

In this unique longitudinal study in which both the effects of a sign language-based intervention and associations between different sign language and cognitive skills and developing reading skills were investigated, we had several predictions that we tested. We predicted that DHH signing children's reading skills would improve over time, and that Omega-is-d2 training would have a positive effect on both word reading and reading comprehension. We also predicted that imitation of unfamiliar signs and sign language PA would be positively related to development of word reading ability, and that sign language comprehension and imitation of familiar signs would be positively associated with development of reading comprehension. For non-linguistic working memory, we predicted positive associations to development in both word reading and reading comprehension.

Methods

Participants

All five Swedish state primary schools for DHH children were invited to take part in this study; two accepted this invitation, and participants were recruited from these schools. Inclusion criteria for the present study were: being at a word reading level corresponding to Grade 1 of hearing children and using Swedish Sign Language (SSL); and, having a hearing impairment (HI). The reason for selecting participants on reading level rather than grade or age, was that reading level is the critical aspect for getting positive effects from Omega-is training (Gustafson et al., 2011; Heimann et al., 1995). The initial selection of participants was made by staff members at the schools on the basis of the inclusion criteria. After initial selection, parents were contacted and asked if they wanted their child to take part in the study. None of the families who were approached declined to participate, and participants and their parents provided informed consent attested in writing by the parents. The study was approved by the Regional Ethical Review Board in Linköping, Sweden. After inclusion in the study, the reading level of the children was assessed on a standardized task, that is, Wordchains (Jacobson, 2001).

Sixteen children (8 boys/8 girls) with a mean age of 10.1 years (*SD* = 2.1) participated, representing 21% of all pupils from Grades 1–7 at the participating schools. Word reading skills of

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the sample assessed 4 months after the start of the academic year did not differ from those of Grade 1 hearing children assessed at a similar point in the academic year (Holmer et al., 2016a). Demographics are presented in Table 1. Three families did not provide full background data. Participants attended grades 1–7. The wide spread in grade level was expected given that it is known that there is high variability in reading skills of DHH signing children (Svartholm, 2010; Qi & Mitchell, 2012). Three of the participants had an additional developmental or medical diagnosis. Two of these and one further participant had corrected vision deficits. All participants had HI and 13 used technical aids (see Table 1). This reflects the fact that although technical aids in many cases have positive effects on speech development in DHH children (Geers & Hayes, 2011; Nakeva von Mentzer et al., 2014), some DHH children with technical aids rely on sign language for communication and learning (Campbell, MacSweeney, & Woll, 2014) and thus attend schools where teaching takes place in sign language. The mean age of fitting of technical aids among the 12 participants for whom data was available was 3.9 years (SD = 2.2). Seven of the participants were born abroad, and the age at which residence in Sweden commenced among the five out of these seven participants for whom data was available ranged from 2:2 to 10:7 years. Non-verbal cognitive ability was screened with Raven's Coloured Progressive Matrices (Raven & Raven, 1994); the three participants with additional disabilities and one further participant performed below the 5th percentile (M = 21.9, SD = 9.0). Participants performed at chance on a measure of Swedish phonological awareness (Holmer et al., 2016a): d' mean score 0.14 (SD = 0.64), $t(15) = 0.84$, $p = .41$, indicating weak Swedish proficiency. The languages spoken in the participants' homes were SSL (n = 5), Swedish (n = 6) or another spoken language (n = 4); data was missing for one participant.

Predictor Variables

Sign Language Skills

Three tasks were used to assess sign language skills. Processing of semantic and phonological representations in the manual modality was assessed using an imitation task (Holmer et al., 2016b) and sign language phonological awareness (PA) was assessed using the Cross-modal Phonological Awareness Test (C-PhAT; Holmer et al., 2016a). Finally, SSL comprehension was assessed with The SSL Receptive Skills Test, an adaptation of a BSL original (Herman, Holmes, & Woll, 1999). The imitation task and C-PhAT were administered with presentation software DMDX (version 4.1.2.0; Forster & Forster, 2003).

Table 1. Demographics

	Group 1 n	Group 2 n
Gender (boy/girl)	4/4	4/4
At least one deaf parent	2	2
Born in Sweden	5	4
Additional disabilities		
Visual (corrected)	1	2
Developmental	2	1
Communication language in school		
SSL	6	7
SSL and Swedish	2	1
Technical aids		
HA	3	3
CI	2	4
HA and CI	1	0
No aids	2	1
Educational level of mother		
Post-secondary	3	2
Secondary or lower	3	5
Unknown	2	1

Note: SSL = Swedish Sign Language; HA = hearing aid; CI = cochlear implant.

Imitation Task

Video clips of a deaf native signer performing different manual gestures were presented on a computer. Three types of gestures were used: familiar lexical forms (i.e., signs from SSL), bearing both semantic and phonological information (i.e., invoking vocabulary skills); unfamiliar lexical forms (i.e., signs from British Sign Language, BSL), bearing phonological information but no meaning; and non-signs, that were gestures that bore no semantic and only reduced phonological information (for more information, see Holmer et al., 2016b). For the present study, only the two first categories were analyzed. Participants saw three videos from each of the three categories on a computer screen (nine videos in all), presented in random order one after the other. After each video the participant was told "Now, it is your turn". If the participant did not attempt to imitate the gesture in the target video, the instruction was repeated once. If there was still no response, the test leader moved on to the next video. Test sessions were video recorded and the precision of imitation of each video-recorded gesture was subsequently rated by two independent sign-naïve coders using a visual analog scale (VAS). Sign-naïve coders were used to ensure that scoring reflected precision in performing the gestural act across all three types of signs. Raters with knowledge of a sign language (e.g., SSL) are likely to be biased by their own gesture-based representations (Rudner et al., 2016). The VAS was a horizontal line on a sheet of paper with fixed end points, marked with "No correspondence" at the left and "Perfect correspondence" at the right. The precision of each sign imitation was rated by marking the VAS with a corresponding cross. The dependent variable was the proportion of the total line between the cross and the left-hand end of the line for each gesture type, averaged across raters. Intraclass correlation coefficients were > .80 for both scores used in the present study, indicating satisfactory reliability.

Cross-modal Phonological Awareness Test

Pairs of printed characters including either one uppercase letter and one digit or two uppercase letters were presented on a computer screen and the task was to determine whether their signed labels shared a single handshape (Holmer et al., 2016a). The Swedish manual alphabet and manual numeral systems are mono-manual and there are a number of instances of handshapes reoccurring across the signed labels of letters and digits (see an example in Figure 1), and this characteristic is utilized in the C-PhAT. Accurate task performance requires the participant to recode the printed digits and letters to their manual equivalents and determine whether the resulting representations

Figure 1. The handshapes for letter Q and digit 6 from the Swedish manual alphabet and manual numeral systems.

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share a single handshape, irrespective of orientation. This task is analogous to determining whether the English labels of, for example, the letter "D" and the digit "3" rhyme. In total, there were 24 trials, consisting of presentation of twelve unique pairs, each presented twice, balancing the order of the printed characters. The pairs are evenly distributed across three categories. In the first category of pairs, the corresponding handshapes of the letters and digits were the same in the Swedish manual alphabet or manual numeral system. C-PhAT can also be used to assess Swedish PA and therefore the Swedish labels of one third of the stimulus pairs rhyme. To avoid confounding effects of Swedish phonological representation, this category of pairs were excluded from the analysis. In the last category of pairs, the labels according to the manual numeral systems were not the same, nor did the Swedish labels of the printed characters rhyme. Since analysis in the present study is based on the first and the last categories of stimulus pairs, only 16 out of the total of 24 trials were used. By pressing one button for yes and one for no, participants had to indicate their responses. The time limit for a response was 20 s and the interstimulus interval was 1 s. Stimulus pairs were organized in blocks, consisting of one pair from each category, and during administration, block order and pair order within blocks were randomized. The number of hits was adjusted for false alarms in accordance with signal detection theory (Swets, Tanner, & Birdsall, 1961), and *d'* was the dependent measure.

Sign Language Comprehension

Participants watched videos of SSL sentences on a computer screen and determined after each sentence which of the three or four alternative line drawings best matched the sentence by pointing to the drawing. The alternatives were visible simultaneously with the video playing, displayed serially in a ring binder placed in front of the screen. A total of 40 sentences were presented. One point was awarded for each correct match and the dependent measure was the total number of points. This test is based on a BSL original for which test–retest reliability has been estimated to .87 (Herman et al., 1999). BSL test scores are associated with expert judgments on the child's signing ability and superior in deaf children of deaf parents compared to children of hearing parents (mean age 8 years, Herman & Roy, 2006). Thus, there is evidence of both satisfactory reliability and validity for this task. The Spearman-Brown split-half reliability estimate from the present data was .96, demonstrating satisfactory reliability. Unpublished data collected 10 months prior to the data collection in the present study were available for two participants and these were used in the present analyses to avoid the additional burden on the individual of retesting. Since no norms are available for the SSL version of this test, performance was assessed in relation to norms for the BSL version for children between the ages of 3 and 11 years (Herman & Roy, 2006). All but two participants were within this age range. Of the four participants with weak performance on Raven's Coloured Progressive Matrices (Raven & Raven, 1994), two scored within ±2 *SD* of the mean according to the BSL norm for their age group, the other two below. The rest of the participants within the age range of the BSL norms, scored either within (n = 8) or above (n = 2) ±2 *SD* of the mean. One participant in the present study was older than 11 years and performed almost 1 *SD* above the mean according to the BSL norm for 11-year olds. Although norms typically vary across different language versions of a test, the comparisons reported here indicate that all but two participants were within the normal range of performance on this test.

Working Memory

The Clown test (Birberg Thornberg, 2010; Sundqvist & Rönnberg, 2010), based on the Mr Peanut task (Kemps, de Rammelaere, & Desmet, 2000), was used as a measure of non-linguistic working memory. In this test, colored magnets were placed at predefined locations on a clown figure attached to a magnetic board. Participants surveyed the configuration for as many seconds as there were magnets. Then the board with the figure was turned away, the magnets were removed and the participant was asked to recall their color. Color recall served to interfere with a language-based rehearsal strategy. After color recall, the participant had to locate the original positions of the magnets by replacing them or pointing. There were ten levels in the task, with an increasing number of locations to be remembered across levels. The first level had one magnet and the last level ten magnets, with one magnet added for each level in between. Each level had three trials, and to pass at a particular level the participant had to respond correctly on at least two out of three trials. The participant was awarded a third of one point for each correct trial, and the dependent measure was total score. Scores on the Clown test (Birberg Thornberg, 2010; Sundqvist & Rönnberg, 2010) correlate strongly with scores on the reading span task, an established measure of working memory (Conway et al., 2005), in children who use augmentative and alternative communication (N = 14; Sundqvist & Rönnberg, 2010). Further, children of a similar age to those in the present study but with difficulties in attentional skills reveal lower scores on this task than typically developing age-matched controls (N = 36; Birberg Thornberg, 2010). These findings suggest that the Clown test is a valid measure of working memory.

Reading Skills

Word Reading

Two tests were used to assess word reading.

Wordchains

In Wordchains (Jacobson, 2001), the participant was presented with uninterrupted strings of written characters (i.e., wordchains) that could be separated into three different Swedish words (e.g., "katt|fot|bil", cat|foot|car). The participant had 2 min to separate using pen strokes as many words as possible in 60 different wordchains evenly distributed in three columns on a sheet of paper. The participant was told to read silently and work as fast as possible. Three additional chains were used for practice before the administration of the test. Wordchains (Jacobson, 2001) is an established test of written word reading in the Nordic countries (Svensson, Lundberg, & Jacobson, 2003), and is also available in English (Miller-Guron, 1999). Importantly, it is commonly used as a measure of word reading for DHH signing children in Sweden. In support of the validity of the task, performance is typically weaker in children with delays in reading development than in typically developing readers (Fälth et al., 2013; Jacobson, 2001). Further, the validity of the task is also supported by its similarity to the parsing of compound words which are common in Scandinavian languages. Test–retest reliability has been estimated to .89 (Jacobson, 2001). The dependent measure was number of chains correctly completed within the time limit.

Lexical decision

In a lexical decision task participants were presented with 40 strings of three lowercase letters on a computer screen in presentation software DMDX (version 4.1.2.0; Forster & Forster, 2003). The strings were 20 real Swedish words, 10 letter strings

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with no meaning in Swedish but that were orthographically legal (i.e., pseudo words), and 10 letter strings that were orthographically illegal in Swedish (i.e., non-words). Items were presented one at a time on the screen, and the task was to judge for each item if it was a real Swedish word or not and appropriately press one button corresponding to yes or another to no. The participant had 5 s to give a response, and the interstimulus interval was 1 s. The dependent measure was *d'* (Swets et al., 1961). Test–retest (between first and second testing) correlation was .71 in the present data, indicating satisfactory reliability. The two tests of word reading were converted into normal scores based on Grade 1 hearing children's performance, thus, a score of 0 would represent mean performance of Grade 1 hearing children (*SD* = 1). The two normal scores were then averaged into a single variable, which was defined as a word reading index. Data from typically developing Grade 1 hearing children were retrieved from norms for Wordchains (*N* = 912; Hogrefe Psykologiförlaget, 2010) and from data collected within this project (*N* = 36; Holmer et al., 2016a) for the lexical decision task.

Reading Comprehension

Reading comprehension was also assessed using two tests.

DLS bas

In DLS Bas (Järpsten, 2004) the participant had to match passages of written Swedish with pictures. For each of the 20 passages, the participant chose which line drawing out of five alternatives best matched the content of the passage by marking the picture with a cross. The first three items are three word sentences (e.g., the first written item is: This is Elin, and apart from the target line drawing showing a girl, the four other drawings show: one boy, one man, two girls and finally one boy and one girl). The passages increase in complexity by including additional word classes, for example, prepositions and adjectives, and sometimes they consist of two sentences (e.g., one of the most complex items is: Elin and her two friends go out onto the balcony. It is windy outside.). Together, the passages create a short story about Elin and her two friends (a tiger and a crocodile). Before testing commenced, two practice items were administered. The participant then had 7 min to silently read the passages and select the most appropriate picture for each passage by marking it with cross. Pictures available for selection with the more complex passages included lures that could only be discounted by detailed understanding of the actors and objects and the relations between them described in the passages. The dependent measure was number of correct answers within the time limit. Test–retest reliability has been estimated to a satisfactory level at .78 (Järpsten, 2004). In support of the validity of the test, scores have been reported to be related to writing ability (Järpsten, 2004). Like Wordchains (Jacobson, 2001), DLS Bas is used to assess reading skills in DHH signing children in Sweden.

Woodcock passage reading comprehension

A Swedish version of the Woodcock Passage Reading Comprehension (WPRC; Woodcock, 1998) test was used (Furnes & Samuelsson, 2009). Text passages with one word omitted, were presented and the task was to say, sign or write a word that correctly completed the passage. There is a progression in difficulty over the 68 items of the Swedish version of WPRC. The first four passages consist of a single sentence of between three and seven words (including the omitted word). Most of the passages in the first half of the test are presented together with a picture that provides a clue to the missing word. By the end of the test, passages involve up to three sentences with both principal and subordinate clauses. In accordance with the recommended test procedure, testing stopped if the participant committed six consecutive errors. The dependent measure was the total number of semantically correct answers. Previous work has shown that the Swedish version of WPRC is positively associated with word reading and rapid automatized naming. This applies to hearing children in Grade 1 (Furnes & Samuelsson, 2009), hearing beginning readers (Fälth et al., 2013), and DHH children who are beginning readers (Nakeva von Mentzer et al., 2014). Further, DHH children who display reading delays perform less well on this task than typically developing hearing children (Nakeva von Mentzer et al., 2014). Test–retest (between first and second testing) correlation in the present study was .77, indicating satisfactory reliability. The two tests of reading comprehension were combined into a reading comprehension index by normalizing raw scores on each test in relation to the performance of hearing children attending Grade 1 and then averaging across tests. Performance of typically developing hearing children attending Grade 1 was derived from norms for DLS Bas (*N* = 248; Järpsten, 2004) and from Furnes and Samuelsson (*N* = 576; 2009) for WPRC.

Omega-is-d2

Omega-is-d2 is a computerized sign language-based literacy training program that, like its forerunners, includes two types of exercises: Create and Test. In the Create exercises (see an example in Figure 2), users produce their own sentences by selecting words or phrases from a list, or set of lists, that appear as columns on the computer screen, and then the semantic content of the sentence is presented as an animation. Further, the user does not have to be familiar with the written words in the lists or the order in which they can be combined; each column corresponds to a fixed position in the type of sentence that can be produced (see Figure 2), and all possible combinations of words within an exercise make up grammatically correct sentences. For example, in a subject-verb-object (S-V-O) sentence, if the user clicks the words in a V-S-O order, the program still displays the correct S-V-O sentence produced by those words. The SSL equivalent of each word or phrase is automatically displayed as a video, directly following its selection, and when the sentence is complete its SSL equivalent is also presented. It is important to note that the complete SSL sentence is not simply the verbatim translation of each of the written words strung together with Swedish word order but the correct propositional equivalent in SSL. Thus, the user is provided with the meaning of the written language string in three different modalities: via the association between the individual written words, that may or may not be correctly identified by the user, via a language form that is familiar to the user (i.e., SSL), and via the animation depicting the meaning of the full sentence. This exemplifies the top-down, or comprehension focused, nature of Omega-is. During the Test exercises, the user sees an animation on the computer screen and then constructs a sentence to match the animation (see an example in Figure 3). When the user correctly selects words (in the correct order) that form the sentence consistent with the animation, positive feedback is given in the form of a further animation (not specifically related to the sentence) but with positive connotations such as an opening flower or a sunrise. However, if the selected words do not form the sentence corresponding to the animation, no feedback is given. Time spent on the exercises is automatically tracked in the program, and can be accessed by the researcher from an administration section.

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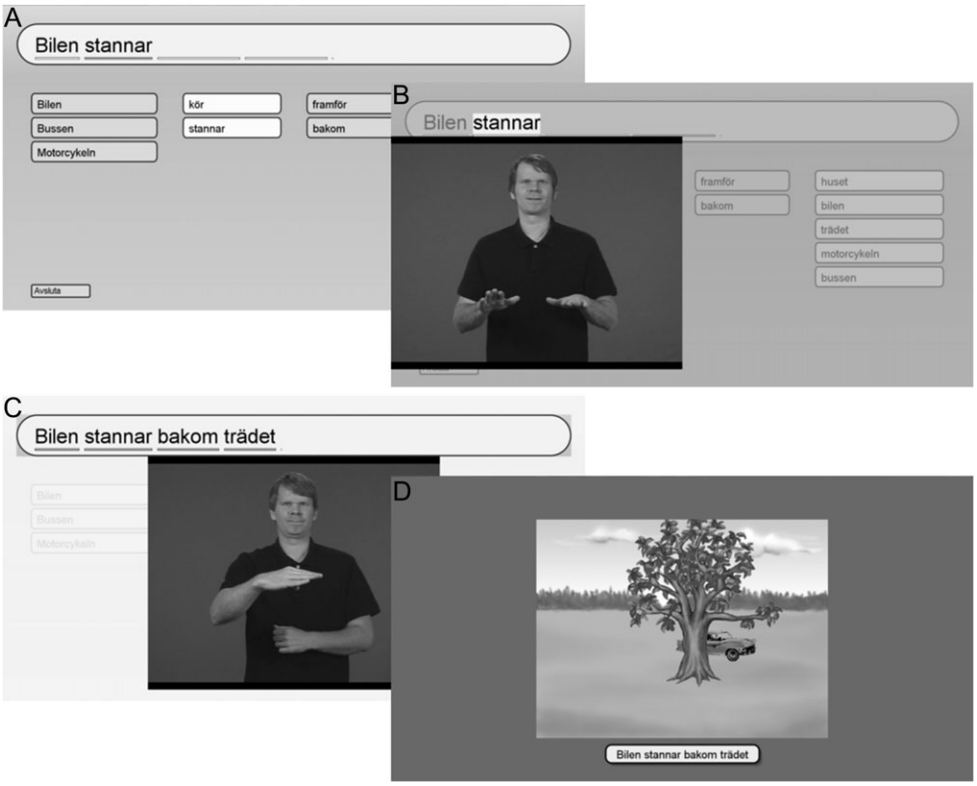

Figure 2. Omega-is-d2: Create. (A) Swedish word "stannar" (*stops*) selected by the user, (B) the Swedish Sign Language (SSL) equivalent for the Swedish word, (C) Swedish sentence, "Bilen stannar bakom trädet" (*The car stops behind the tree*), has been created by the user and the SSL equivalent of that sentence is presented on the screen, (D) finally the sentence appears as an animation, with the printed version of the sentence displayed below. Reproduced with permission from the actor and the licenser.

The generic Omega-is has several levels: the first level involves single nouns (e.g., The penguin); at the second level, nouns and verbs can be combined into two word subject-verb sentences (e.g., The frog jumps); the third level also involves nouns and verbs, which are used for three word sentence Swedish (e.g., ["Lejonen jagar svanen"] "The lion chases the swan"); at the fourth level, prepositions are added (e.g., The panda puts the pizza on the whale's table); and in the fifth, and last level, conjunctions and adjectives are also included (e.g., The singing fox runs over the bridge and hugs the bear). In addition to these different levels of the program, it also includes a part where shorter stories can be constructed following steps in which different words and phrases are combined. For our earlier sign-based version, Omega-is-d1 (Rudner et al., 2015), only the first three levels of Omega-is were translated into SSL and the animations had to be dropped for technical reasons. In the current version, Omega-is-d2, all text material from the generic Omega-is version, except the short stories, was translated into SSL and video recorded. Thus, the amount of material that the participant could work with was substantially larger than in our previous study (Rudner et al., 2015), keeping the content interesting to work with for participants who progress beyond three word sentences during training. In total, more than 1,700 SSL videos were recorded for the present version, in comparison to 220 videos in our earlier study (Rudner et al., 2015). Because the SSL sentences follow SSL grammar and are not merely serial concatenations of the signs corresponding to the words or phrases in Omega-is, all possible full length sentences from Omega-is as well as each individual word and phrase were video filmed and incorporated in Omega-is-d2. Materials were produced in collaboration with the Sign Language Section of the Department of Linguistics, Stockholm University, and two deaf native signers. The animations from the original program were maintained in Omega-is-d2.

There are several aspects of the design of Omega-is-d2 that are likely to reduce working memory load and thus release cognitive resources for learning. Firstly, the level at which training starts is the level at which the participant gets four out of five sentences correct in the Test exercises. This means that the intervention starts at an appropriate linguistic level for the individual participant. Further, in the Create exercises, the participant determines the order in which each word is connected to its equivalent sign, and at the pace at which sentences are created. After each a word has been clicked on and highlighted, or a sentence has been constructed (see Figure 2), there is also a short time lag before presentation of the corresponding sign. These aspects are likely to facilitate temporal processing and reduce the load on attentional resources, allowing the participant to focus on integrating the information that is presented on screen. On top of this, the program supports processing by providing a structure for combining words into grammatically correct sentences. This helps the participant to build larger meaningful chunks (i.e., sentences) from smaller meaningful components (i.e., words). This form of controlled segmenting on the user side, and supportive structure on the software side, lead to better learning from multimedia (Mayer & Moreno, 2003). Another design feature that is likely to reduce working memory demands by reducing demands on visual processing, is that sign equivalents and target written word are presented in close proximity to each other (Mayer & Moreno, 2003). Visual

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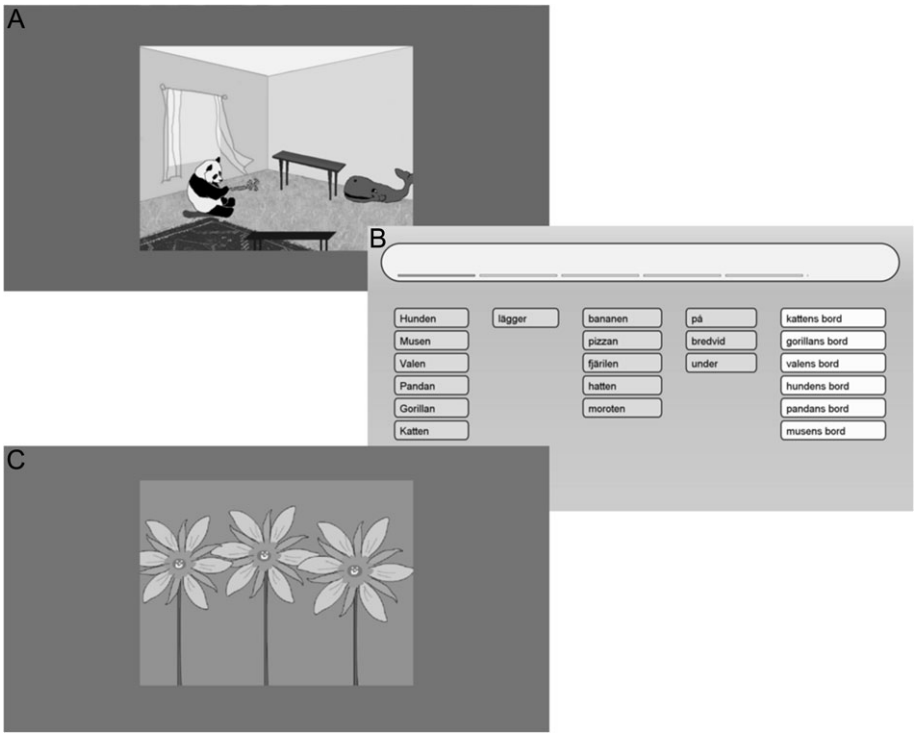

Figure 3. Omega-is-d2: Test. (A) An animation is presented without any associated language material, (B) from a restricted set of words and phrases, the user creates a sentence that correctly describes the animation, (C) if the user creates a correct sentence, visual feedback is provided before a new animation appears, and otherwise a new animation is displayed directly after the response. Reproduced with permission from the licenser.

processing demands are further reduced by a visual filter that overshadows extraneous visual information (i.e., other written words) when a word is selected. Decreasing extraneous information, which otherwise has to be suppressed, has also been shown to increase learning from multimedia interventions (Mayer & Moreno, 2003). Although there are situations in multimedia learning when static depiction of events is preferable to dynamic (i.e., animations), Höffler and Leutner (2007) concluded based on a meta-analysis that dynamic presentation produced better learning outcomes than static as long as presentation only included target-relevant information. This is achieved in Omega-is-d2, since the activity in the animations simply represents the meaning of the sentence, thus avoiding noise induced by irrelevant information. In addition, animations are presented simultaneously with the written sentences, which means that the learner does not need to rely on working memory to make the comparison (Mayer & Moreno, 2003). Last, but not least, recasting, that is, changing an utterance by the addition, deletion or permutation of information, while maintaining its meaning (Bohannon, Padgett, Nelson, & Mark, 1996), supports maintained attention to relevant information and decreased working memory demands, since meaning is repeated in several different forms (i.e., written language, sign language and animation) within a short time period (Nelson et al., 1997). Providing meaning externally allows cognitive resources to be used for analyzing the connection between language forms and meaning rather than retrieving representations from long-term memory.

In the present study, training took place in school and participants were encouraged to work with the program for at least 10 min per day over a period of 4 weeks (20 days, Fälth et al., 2013; Gustafson et al., 2011). Participants were free to work with both Create and Test exercises to whatever extent they chose, but were encouraged to focus on the former type of exercises since these were designed to established connection between signed and written language. Whenever possible without neglecting their regular duties, teachers sat next to the participants and supported training by, for example, initiating discussions on differences in syntactic structure across sign language and written language.

	Test occasion				
		Cross-over period			
	T1 Week 0	T2 Week 5	T3 Week 10	T4 Week 16	T5 Week 39
Assessment	Sign language skills				
	Working memory				
	Reading skills	Reading skills	Reading skills	Reading skills	Reading skills

Figure 4. Study design.

Procedure

To investigate development over time and test the effects of Omega-is-d2 training on reading skills, a longitudinal cross-over design was implemented. To avoid potentially unmatched experimental groups generated by a randomization procedure, given the small sample size, participants were divided into two groups that were matched on several background variables (see Table 1). Age, $t(14) = 1.01$, $p = .33$, and distributions of gender, country of birth and additional medical or developmental disabilities were similar across groups. Word reading and reading comprehension were assessed at five occasions (T1–T5), that is, at 0, 5, 10, 16, and 39 weeks (see Figure 4). The relatively longer time spacing between T3 and T4 (5 weeks instead of 4 weeks), was due to the fact that participants were on school holiday for

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1 week during this period. The follow-up at T5 was placed 9 months after the start of the study, corresponding to approximately 6 months after the end of the cross-over period. Between Week 5 and Week 16 (T2–T4) the cross-over was executed. Group 1 received Omega-is-d2 training from T2 to T3, and Group 2 from T3 to T4. The order in which the two groups received training was randomized. Predictor variables were assessed between the two first test occasions, before Omega-is-d2 training commenced. Some of the data relating to the predictor variables and the reading tasks at the initial and final test occasions have been included in other analyses reported elsewhere (Holmer et al., 2016a, b, Holmer, Heimann, & Rudner, 2016c).

To test our predictions, while at the same time dealing with dependency and unequal time spacing between test occasions, we deemed hierarchical linear models (HLMs) to be the most appropriate procedure. HLMs are more flexible than traditional approaches to longitudinal data analysis (e.g., analysis of variance), and can effectively handle missing data, dependency and unequal time spacing between test occasions (Hesser, 2015; Singer & Willett, 2003). Further, HLMs allow inclusion of predictors of change in the dependent variable, as well as predictors that are both time-varying (i.e., predictors that can change over time, such as receiving or not receiving an intervention) as well as time-invariant (i.e., predictors that have a fixed value, such as level of a specific skill at a first test occasion). However, although parameter estimates can be unbiased when running HLMs on small samples, variances may be underestimated (Maas & Hox, 2005), leading to an increased risk of Type I errors. Others have suggested that HLM is a useful strategy for analyzing longitudinal data from small samples, although results should be seen as indicative rather than definitive (Davis et al., 2013). The hazards of applying HLM on small samples are taken into account in the interpretation of results.

Firstly, descriptive statistics for predictor variables and measures of reading skills across the five test occasions (T1–T5; see Figure 4) were calculated and group comparisons were conducted. In a second step, HLMs were fitted to the word reading index (WR) and the reading comprehension index (RC) to investigate change in reading skills over time (T1–T5) and effects of Omega-is-d2 on developing reading skills (fitting of HLMs is described below). Thirdly, predictors (C-PhAT-SSL, Imitation of unfamiliar signs, Imitation of familiar signs, SSL comprehension, and Working memory) were added to the two final HLMs (one for WR and one for RC) from the second step, to investigate whether any of them could explain rate of change in WR or RC. To restrict the number of models tested on our small data set, modeling was based on our theoretically driven predictions rather than patterns in our data. That is, for WR, only C-PhAT-SSL, Imitation of unfamiliar signs, and Working memory were used as predictors, and for RC, only SSL comprehension, Imitation of familiar signs, and Working memory. All statistical computations were conducted in IBM SPSS Statistics (Version 22.0).

Missing Data

One participants did not do the SSL comprehension test. Due to technical errors, all responses on the imitation task were missing for two participants, and one further participant had one missing response on Imitation of unfamiliar signs.

For the reading measures, 14% and 11% of the data points were missing in total from T1, T2, T4, and T5 (no data was missing at T3). For word reading, results from both measures were missing for three participants at T1, and from six children at T2. At T4 and T5, results from one participant were missing. In addition, two participants only had results from one of the two word reading measures at T1 and at T2. In these two cases, the one available data point was used for WR.

For reading comprehension, results from both measures were missing for three participants at T1, and from four and at T2. At T4 and T5, the result from one child was missing. In addition, four participants at T1 and T2 plus one at T4 only had results from one of the two reading comprehension measures. For these children, the one available data point was used for RC.

HLMs provide unbiased estimates if data is missing under the MCAR (missing completely at random) or MAR (missing at random) mechanisms (Enders, 2010). However, in small samples missing data can further increase the risk of inflated Type 1 errors (McNeish, 2017), which warrants even more caution when interpreting results. Given that missing data was mainly due to technical errors in the present study, a MAR mechanism was assumed for all missing data. Further, adding variables that are associated with the probability of missingness to HLMs reduces bias in the model (Enders, 2010). More data was missing from one of the participating schools, and thus a dummy variable (i.e., context) was used in a final step of modeling to control for any confounding effects of this.

Hierarchical Linear Modeling

By incorporating individual variation (i.e., random effects) around fixed effects, HLMs can effectively handle dependency of data in repeated-measures designs (Hesser, 2015; Singer & Willett, 2003). As a first step in the present study, individual and group scatterplots were visually inspected to examine how to model data. In a second step, unconditional growth models were constructed for each of the two reading indices. Unconditional growth models only include a time variable as the independent variable and the repeated measure (in this case WR or RC) as the dependent variable, and may or may not include random intercepts (i.e., individual variability at time 0) and/or random slopes (i.e., individual variability in change slopes) (Singer & Willett, 2003). The unconditional growth model for WR was set to be linear over time with uncorrelated and constant residuals (identity covariance structure for the repeated measure), and individuals were allowed to vary in both their intercepts (i.e., random intercepts) and slopes (i.e., random slopes). The unconditional growth model for RC was set to be curvilinear over time, since data indicated no change in RC between T4 and T5, had uncorrelated and constant residuals, and individuals had random intercepts. To test the effect of Omega-is-d2 training, a time-varying intervention variable, that is, coded as 1 for each occasion following an intervention period and as 0 for all other occasions, was added to each of the two unconditional growth models. One participant did not receive training due to absence from school. This participant was still included in modeling, and the intervention variable was set to 0 at all occasions (excluding this participant from analysis did not change the overall pattern of results). Finally, to investigate which predictor variables were related to development in word reading and reading comprehension, several conditional growth models were tested (all predictor variables were centered). Due to lack of space, only a few models are reported in the results, but all models, their fit indices, variance structure, and fixed and random parameters are described in Appendix A (WR) and Appendix B (RC). Although restricted maximum likelihood estimation is recommended when fitting HLMs on small data sets, models were fitted using full maximum likelihood (ML) estimation so that deviance statistics (i.e., difference in −2*Log likelihood between models) could be used

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to test whether a new model (e.g., comparing models with and without the intervention variable) was a better fit to data than a preceding one (Singer & Willett, 2003). However, for the final models both estimation methods were compared. Results for the fixed effects, which were of primary focus in the present study, were similar across methods and models built with ML estimation are reported.

Results

Participants worked with Omega-is-d2 for an average of 12 days (*SD* = 5) during a 4-week intervention period, and used the program on average for 33 min (*SD* = 24) per day, which was more than three times what we recommended. Time per day spent on Create was on average 18 min (*SD* = 14) and on Test on average 15 min (*SD* = 22). Groups did not differ on predictor variables or amount of training with Omega-is-d2 (see Table 2). Reading indices over the five test occasions for the two groups are displayed in Table 3. Concurrent associations between reading indices ranged from medium sized to large (*r*'s .43–.86) across assessment points. Inspection of scores on the word reading index (WR) and reading comprehension index (RC) indicated that participants performed worse than mean performance of hearing Grade 1 children on reading comprehension at all assessment points.

Modeling Change Over Time

Results from selected HLMs, including fit indices, that is, deviance statistics (−2 Log likelihood, −2LL), Akaike's Information Criterion (AIC), and Schwartz's Bayesian Criterion (BIC), are reported in Table 4 (WR) and Table 5 (RC). See Appendix A (WR) and Appendix B (RC) for results from all HLMs. Both for WR (see Table 4) and for RC (see Table 5) there was a significant effect of time, indicating that reading skills improved in the group over the five test occasions. For WR, the improvement in model fit by adding the intervention variable was approaching significance, $\chi^2(1) = 3.688$, $p = .055$, indicating a possible effect of the intervention on WR, estimate = 0.21, 95% CI [−0.01, 0.43], $p = .057$ (Model 1 in Table 4). In Figure 5, scores on WR for the two groups across the cross-over period (T2–T5) are displayed. For RC model fit was not improved by adding the intervention variable, $\chi^2(1) = 0.142$, $p = .71$ (Model 1 in Table 5). Thus, Omega-is-d2 training did not have a stronger impact on reading comprehension than regular schoolwork. In Figure 6, scores on RC for the two groups across the cross-over period (T2–T5) are presented. Since the theoretical motivation behind using Omega-is-d2 is to connect existing language representations to new linguistic forms, we tested whether excluding the two participants who performed below the norms for SSL comprehension influenced the pattern of results. However, the pattern was still the same, with no statistically significant effects of the intervention.

Predicting Development in Reading Skills

As predicted, imitation of unfamiliar signs predicted both initial level, estimate = 0.02, 95% CI [0.01, 0.04], $p = .002$, that is, individual variability at Week 0, and development over time, estimate = 0.0004, 95% CI [0.000002, 0.0008], $p = .049$, that is, the rate of change in WR across test occasions (see Model 2 in Table 4). Even

Table 2. Descriptive statistics on predictor variables and amount of training in Omega-is-d2 for the two experimental groups and the full sample

	Group 1			Group 2			Full sample				
Variable	*M*	*SD*	95% CI	*M*	*SD*	95% CI	*M*	*SD*	95% CI	*t*	*p*
C-PhAT-SSL	0.9	1.1	[−0.1, 1.8]	0.9	1.1	[0.0, 1.8]	0.9	1.1	[0.3, 1.5]	0.09	.93
Familiar signs	35.3	15.4	[21.1, 49.6]	57.0	27.6	[31.5, 82.6]	46.2	24.3	[32.2, 60,2]	1.82	.09
Unfamiliar signs	41.4	15.7	[26.9, 55.9]	54.3	26.0	[30.3, 78.4]	47.9	21.7	[35.3, 60.4]	1.13	.28
SSL comprehension	30.3	5.6	[25.1, 35.5]	29.9	12.5	[19.5, 40.3]	30.1	9.6	[24.8, 35.4]	0.08	.94
Working memory	1.8	0.9	[1.1, 2.5]	1.9	0.8	[1.3, 2.5]	1.9	0.8	[1.5, 2.3]	0.21	.84
Omega-is-d2, Days	12.3	6.5	[6.8, 17.7]	10.4	5.8	[5.5, 15.2]	11.3	6.0	[8.1, 14.5]	0.61	.55
Omega-is-d2, min/day	39.5	28.6[a]	[15.6, 63.5]	25.4	15.1	[11.5, 39.4]	32.9	23.7	[19.8, 46.0]	1.16	.27

Note: *T*-test statistics are based on comparison between Group 1 and Group 2. C-PhAT-SSL = Cross-modal Phonological Awareness Test, Swedish Sign Language version; SSL = Swedish Sign Language.
[a]One participant had an extreme time (>100).

Table 3. Scores on word reading (WR) and reading comprehension (RC) indices across the five test occasion (T1–T5) in both intervention groups and the full sample

		Test occasion														
		T1			T2			T3			T4			T5		
Variable	Group	*n*	*M*	*SD*	*n*	*M*	*SD*	*n*	*M*	*SD*	*n*	*M*	*SD*	*n*	*M*	*SD*
WR	1	7	−0.5	0.6	4	−0.5	0.7	8	0.0	0.8	7	−0.3	0.6	7	−0.1	0.5
	2	6	−0.4	0.9	6	0.1	1.1	8	0.2	1.1	8	0.4	1.2	8	0.7	1.4
	All	13	−0.4	0.7	10	−0.1	1.0	16	0.1	1.0	15	0.1	1.0	15	0.3	1.1
RC	1	7	−2.0	0.5	5	−1.5	0.3	8	−1.6	0.4	7	−1.7	0.4	7	−1.6	0.5
	2	6	−1.7	0.5	7	−1.4	0.5	8	−1.3	0.6	8	−1.1	0.6	8	−0.9	0.7
	All	13	−1.9	0.5	12	−1.4	0.4	16	−1.4	0.5	15	−1.4	0.6	15	−1.2	0.7

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Table 4. Parameter estimates, standard errors (SE) and confidence intervals (CI) and model fit indices for selected growth models for the word reading index

	Model 1			Model 2		
Parameter	Estimate	SE	95% CI	Estimate	SE	95% CI
Fixed effects						
Intercept	−0.25	0.20	[−0.67, 0.18]	−0.37*	0.14	[−0.67, −0.07]
Linear slope	0.02**	0.005	[0.01, 0.03]	0.02**	0.004	[0.007, 0.02]
Omega-is-d2	0.21†	0.11	[−0.01, 0.43]	0.22†	0.11	[−0.002, 0.45]
Unfamiliar signs	—	—	—	0.02**	0.01	[0.01, 0.04]
Unfamiliar signs*Linear slope	—	—	—	0.0004*	0.0002	[0.000002, 0.0008]
Random effects	0.13***	0.03	[0.11, 0.25]	0.13***	0.03	[0.10, 0.23]
Intercept	0.58**	0.26	[0.33, 1.43]	0.21*	0.09	[0.10, 0.54]
Slope	0.0002	0.0001	[0.00005, 0.0006]	0.0001	0.0001	[0.00001, 0.0006]
Model fit indices						
Repeated, Pseudo R^2	.08			—		
Intercept, Pseudo R^2	—			.64		
Slope, Pseudo R^2	—			.54		
−2LL	114.314			87.822		
AIC	126.314			103.822		
BIC	139.719			120.967		
χ^2 (Change in −2LL)	3.688, df = 1, p = .055			26.492, df = 2, p < .001		

†p < .10. *p < .05. **p < .01. ***p < .001.

Table 5. Parameter estimates, standard errors (SE) and confidence intervals (CI) and model fit indices for relevant growth models for the reading comprehension index

	Model 1			Model 2		
Parameter	Estimate	SE	95% CI	Estimate	SE	95% CI
Fixed effects						
Intercept	−1.69***	0.13	[−1.96, −1.42]	−1.76***	0.09	[−1.95, −1.58]
Linear slope	0.03*	0.01	[0.002, 0.06]	0.03*	0.01	[0.007, 0.05]
Quadratic slope	−0.0005	0.0003	[−0.001, 0.0002]	−0.0005†	0.0003	[−0.001, 0.00003]
Omega-is-d2	0.05	0.12	[−0.19, 0.28]	—	—	—
Familiar signs	—	—	—	0.01**	0.003	[0.004, 0.02]
Familiar signs*Linear slope	—	—	—	0.0002†	0.0001	[−0.00002, 0.0005]
Random effects	0.11***	0.02	[0.08, 0.16]	0.10***	0.02	[0.07, 0.15]
Intercept	0.18*	0.07	[0.08, 0.40]	0.03	0.02	[0.01, 0.12]
Model fit indices						
Repeated, Pseudo R^2	.00			.09		
Intercept, Pseudo R^2	—			.82		
−2LL	78.968			48.648		
AIC	90.968			62.648		
BIC	104.544			77.869		
χ^2 (Change in −2LL)	0.142, df = 1, p = .71			30.320, df = 2, p < .001		

†p < .10. *p < .05. **p < .01. ***p < .001.

though C-PhAT and Working memory predicted initial level when used as individual predictors in two separate models, both associations lapsed when imitation of unfamiliar signs was included as a second independent variable in addition to each of these predictors (see Appendix A). For RC (see Model 2 in Table 5), imitation of familiar signs predicted initial level, estimate = 0.01, 95% CI [0.004, 0.02], p = .003, and had a marginally significant effect on the linear slope, estimate = 0.0002, 95% CI [−0.00002, 0.0005], p = .073. This pattern of results was the same when excluding the two participants with below typical performance on SSL comprehension. SSL comprehension predicted initial level when used as the only predictor of RC, but this association disappeared when both SSL comprehension and imitation of familiar signs were included as predictors (see Appendix B).

Discussion

In the present study, both word reading and reading comprehension improved significantly over a 9-month period in DHH signing children who were at an early stage of reading development. Further, results indicated that rate of change both in word reading and in reading comprehension were associated with sign language skills. In particular, development in word reading was predicted by precise imitation of unfamiliar signs. Further, the improvement in model fit achieved by adding the

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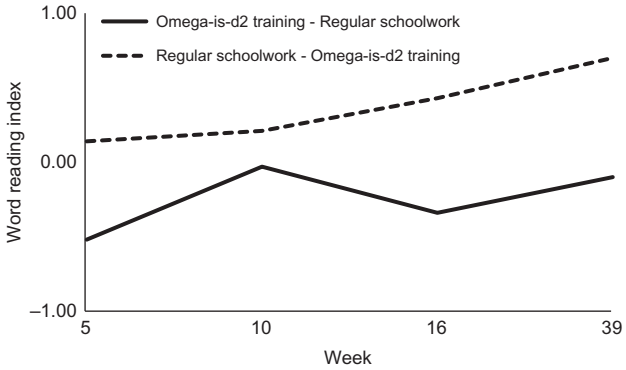

Figure 5. Development in word reading index for Group 1 (Omega-is-d2 training followed by regular schoolwork; dotted line) and Group 2 (regular schoolwork followed by Omega-is-d2 training; solid line) during the cross-over period (Week 5–Week 16), and to the follow-up (Week 39).

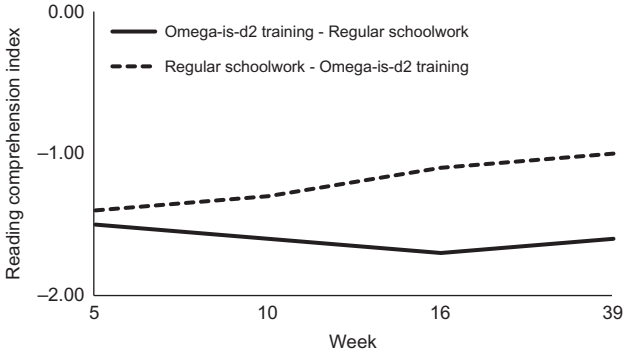

Figure 6. Development in reading comprehension index for Group 1 (Omega-is-d2 training followed by regular schoolwork) and Group 2 (regular schoolwork followed by Omega-is-d2 training) during the cross-over period (Week 5–Week 16), and to the follow-up (Week 39).

Omega-is-d2 intervention variable was marginally significant for word reading but negligible for reading comprehension. Thus, no significant effect of the computerized sign language-based intervention, Omega-is-d2, could be established on either word reading or reading comprehension skills. In addition, the predicted associations between reading development and the three variables sign language phonological awareness, sign language comprehension and non-linguistic working memory were not significant.

Sign Language Skills and Developing Reading Skills

Results of the present study show that participants who imitated with higher precision gestures that bore a high degree of phonological information, that is, unfamiliar signs, had a steeper development in word reading. The imitation task used here is a novel approach for assessing sign language skills, and although Marshall (2014) argues that it is cognitively equivalent to similar tasks in spoken language, this has yet to be confirmed empirically. Nevertheless, this finding is a replication of earlier findings reporting concurrent connections between sign language skills and reading skills in DHH signing children (Hermans et al., 2008b; McQuarrie & Abbott, 2013; Schönström, 2010), with the added value of being longitudinal. However, it should be interpreted with caution, given the small and heterogeneous sample and a relatively high degree of missing data (Davis et al., 2013; Maas & Hox, 2005; McNeish, 2017).

Imitating unfamiliar signs and learning to decipher the orthographic representations of words both involve the active maintenance of a new surface form in working memory during comparison with prior representations (Holmer, 2016). This ability may indicate sensitivity for change in the lexical system, and might be a particularly important part of word reading development for DHH signing children, because these children need to establish new language representations when learning to read (Hoffmeister & Caldwell-Harris, 2014; Svartholm, 2010). In contrast to our predictions, on the other hand, non-linguistic working memory capacity and sign language phonological awareness (PA) did not predict rate of change in word reading. However, change in the lexical system is likely to be a result of the interaction between working memory capacity and phonological representations (Gathercole, 2006; Metsala, 1999). Even though both may be necessary preconditions in word reading development, the interaction between short-term storage and long-term representations might be of particular importance when DHH signing children are learning to read words. We tentatively suggest that one critical component might be the explicit use of stored sub-lexical representations in new ways, which leads to a restructuring of the lexical system (e.g., establishment of new lexical representations; Holmer, 2016).

Regarding reading comprehension, there was a marginally significant association between rate of change and precision of imitation of familiar signs. This is in line with earlier reports of connections between vocabulary and reading comprehension in DHH signing children (Hermans et al., 2008b), as well as a study by Gentry et al. (2004/2005) in which semantic cues (i.e., pictures) aided comprehension of texts in deaf children. Further, the present finding also concurs with findings from deaf adults (Hirshorn et al., 2015), and the notion that DHH signing children can use their sign vocabulary to access meaning of written words at early stages of reading development (Crume, 2013; Haptonstall-Nykaza & Schick, 2007; Hermans et al., 2008a; Hoffmeister & Caldwell-Harris, 2014). In addition, both reading specific (Language and Reading Research Consortium, 2015; Perfetti & Stafura, 2014) and language general (Kintsch & Rawson, 2007; Rönnberg et al., 2013) models of comprehension, suggest that semantic processes related to vocabulary are crucial for language understanding. With only a marginally significant effect and the issues associated with the characteristics of the present data set, the association between rate of change and precision of imitation of familiar signs should be interpreted with caution. However, it is safe to say that for DHH signing children who are learning to read, semantic processes relating to sign language might support the development of reading comprehension (Holmer, 2016). Thus, sign vocabulary may play a central role in development of reading comprehension in DHH signing children, and focusing on establishing a rich sign vocabulary during the years before formal reading instructions begins might provide a firm foundation for reading development in DHH children (c.f., Lederberg, Schick, & Spencer, 2013). However, the role of vocabulary in developing reading comprehension needs to be studied in larger samples, to determine whether the preliminary findings in the present study can be replicated and extended.

Contrary to our predictions, non-linguistic working memory and sign language comprehension did not predict development in reading comprehension. According to the ELU model (Rönnberg et al., 2013), language processing builds on

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maintenance and updating of a representational model as new information enters the system. Under adverse processing conditions, updating is hindered and language understanding constrained by individual working memory capacity. However, we found no evidence of such constraint in our data. The nonlinguistic working memory task used in the present study was selected to be independent of a specific language modality (spoken, signed or written). One possibility is that the visuo-spatial nature of the task tapped abilities that are not reflected in reading comprehension. Another possibility is that working memory has to reach some threshold maturity, not yet achieved by the participants in the present study, before it is even possible to begin to construct representational models of written language.

Although sign language skill has been found to be positively related to reading comprehension (Chamberlain & Mayberry, 2008; Schönström, 2010; Stone et al., 2015), it has been suggested that even though sign language comprehension is necessary for DHH signing children to begin to learn to read, comprehension skills do not automatically transfer across language modalities (Goldin-Meadow & Mayberry, 2001; Holzinger & Fellinger, 2014). Rather, sign language comprehension sets a baseline from which reading comprehension can emerge, but it cannot do so without adequate exposure to print and support from tutors who are more knowledgeable. However, there are also some limitations to consider when interpreting the lack of association between sign language comprehension and reading comprehension development in the present study. The sample here was at an early stage of their reading development, and the results on reading comprehension indicated trending floor effects suggesting that true understanding of text content was restricted. At later stages of reading development sign language skill may play a different role. Indeed, language comprehension typically becomes more strongly associated with reading comprehension at stages of reading development later than those studied here (Garcia & Cain, 2014; Language and Reading Research Consortium, 2015; Ripoll Salceda et al., 2014). Further, power was restricted in the present study and the relatively small change in reading comprehension over time, limiting the amount of variance to explain, might have confounded results. The selected measures of reading comprehension may also play a part in our results, since different reading tasks are likely to rely more or less heavily on specific cognitive resources (Garcia & Cain, 2014) tapped by the predictors used here. However, investigation of this issue is beyond the scope of the present study. Future studies could utilize the imitation paradigm presented here, in combination with other measures of working memory, sign language skills and reading skills, in larger and less heterogeneous samples, to investigate how the active representation of semantic and phonological information in working memory relates to reading development in DHH signing children.

Although word reading skill of the present sample was more or less on par with typically developing Grade 1 readers across all five test occasions, reading comprehension was well behind. This finding is in line with the proposal that word reading and reading comprehension might be more loosely coupled for DHH signing children than for hearing children (Marschark & Wauters, 2008). A greater separation between word reading and reading comprehension skills in DHH signing children is likely to have to do at least in part with the lack of correspondence between meaningful sign-based representations and orthographic forms. The possibility that word reading and reading comprehension are more loosely coupled in DHH signing children than in hearing children should be further investigated in future studies.

Omega-is-d2 Training

In line with both our initial prediction and the observed positive association between imitation of unfamiliar signs and rate of change in word reading over time, we saw a marginally significant effect of the intervention on word reading. Although a positive effect of the intervention is supported both by theory and empirical evidence, the characteristics of the present data set restricts what interpretations that can be made. In addition, the effect on reading comprehension was negligible. Earlier studies on hearing children, with word reading skills similar to those of the participants in the present study, have reported positive effects of Omega-is on developing reading skills after a period of time comparable to that in the present study (e.g., Gustafson et al., 2011). Further, earlier studies on DHH signing children indicate that interventions aimed at strengthening the connections between sign language and written language may be efficient for learning to read and comprehend new written words (Haptonstall-Nykaza & Schick, 2007; Wauters et al., 2001). Similarly, connecting written words to their meaning via presentation of pictorial material also seems helpful (Reitsma, 2009). In the present study, written words and sentences were connected both to sign language translations and to animations (i.e., pictorial materials). Thus, the lack of assuring effects of Omega-is-d2 is puzzling. On the other hand, few earlier published studies have investigated the effects of reading interventions on reading skills in DHH children who use signed languages, and we have limited knowledge about how reading interventions should be constructed and implemented to reach valuable outcomes for this group. The marginally significant effect on word reading could be one important lead in the search for effective reading interventions in this group, which should be followed up in future studies on sign-based versions of Omega-is.

The present study is to our knowledge the first in which the generalizability effects of a reading intervention for DHH signing children who are learning to read are assessed. It is hard to determine exactly what changes on the reading measures in the present study represent, for example, test–retest, intervention or schooling effects or a mix of these, since the stability in performance on these measures in DHH signing children is not known. However, because there was significant effect of time for both word reading and reading comprehension, the lack of effect of the intervention was not due to our reading measures being insensitive to detect changes. However, there are aspects relating to sample characteristics and implementation of the intervention that might explain why no convincing effects were detected.

Firstly, DHH signing children are not only learning to read in a second language but also a language that is based on a modality different from that of their primary language (Hoffmeister & Caldwell-Harris, 2014; Marschark & Wauters, 2008). The participants in the present study was recruited in a similar matter and from a similar context as the participants in our study on Omega-is-d1 (Rudner et al., 2015), and the characteristics of samples were alike. Perhaps, DHH children who use sign language need to establish a level of language representation, not yet achieved by the participants in the present study (c.f., Rudner et al., 2015), before written language can be meaningfully represented in working memory. Based on earlier studies on Omega-is with hearing children (Gustafson et al., 2011), word reading skill at a Grade 1 level was taken as a marker of whether or not participants were sufficiently mature linguistically for benefiting from training in the present study (c.f., Rudner et al., 2015). However, the qualitative difference across hearing and

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DHH signing children regarding the correspondence between prior representations and written language might produce a shift in the level of initial reading skill participants need to gain from a short-term intervention like the one in the present study. To learn from Omega-is-d2 training, DHH signing children might need a certain level of orthographic (Bélanger & Rayner, 2015) or speech-based (Sehyr, Petrich, & Emmorey, 2017) representation that can be actively held in working memory. A further possibility is that sign language proficiency needs to reach some threshold level before effects of Omega-is-d2 training can be observed. Since the most crucial part of Omega-is-d2 training, as implemented in the present study, is strengthening the connections between sign language and written language, sign language proficiency becomes a bottleneck for learning. Although performance on our measure of Swedish Sign Language (SSL) comprehension seemed to be typical when comparing to British Sign Language norms for all but two participants in the present study, the specific test used here to assess SSL proficiency might not be sufficiently sensitive to detect atypical development of the specific type of sign language skill most crucial for reading development. Based on the characteristics of the sample, it is likely that several participants actually had not had the chance to develop age-appropriate SSL. For example, only five participants primarily used SSL in their homes, and around half of the participants were not born in Sweden and did not receive any SSL before the age of two at the earliest. It is likely that background factors like these influence SSL proficiency (Mayberry, Chen, Witcher, & Klein, 2011; Mayberry, Lock, & Kazmi, 2002), and that this in turn has an impact on literacy development (Lederberg et al., 2013). Indeed, results from a recent large-scale study by Clark et al. (2016), suggests that early access to sign language for DHH signing children leads to better reading outcomes than later access. Thus, although both word reading skills and SSL proficiency seemed to be adequate for profiting from Omega-is-d2 training, there might be linguistic characteristics of the present sample that explain why we did not detect stronger effects of training. The type and level of sign language skills that are needed for benefitting from the type of training involved in Omega-is-d2, and similar interventions, should be investigated in future studies. The finding in the present study of a statistically significant association between word reading and the ability to precisely imitate unfamiliar signs, raises the possibility that it is the ability to represent phototactically acceptable but semantically empty manual gestures that underpins word reading. Such a notion is in line with the proposal by Stone et al. (2015) that fingerspelling ability may be important for decoding written words. Lack of appropriate sign language skills might be one reason why DHH signing children progress slowly, and sometimes not at all, in their reading development (Goldin-Meadow & Mayberry, 2001). An additional note, related to the linguistic preferences of the participants, is that it is possible to include both spoken Swedish and SSL videos in Omega-is-d2. However, for the present study, where the aim was to investigate the effects of connecting sign language to written language, all speech material was deleted from the program. Further, participants performed at chance on a measure of Swedish PA (Holmer et al., 2016a), indicating weak spoken language proficiency and low likelihood of benefiting from speech materials in Omega-is-d2 at a group level. On the other hand, a couple of participants in the present study did use some speech as a complement to sign language and might have benefitted from dual language codes during training. Future studies could try to optimize learning conditions (Nelson, 1998) by tailoring Omega-is-d2 for each user.

Other possible reasons for the lack of convincing effects of the intervention are issues relating to the practical implementation of the training. To begin with, although the intervention ran for 20 days, the average exposure of the sample as a whole to the intervention was not much higher than the 10 days reported from our previous study (Rudner et al., 2015). Further, how much time participants spent in the two types of exercises (i.e., Create and Test) varied. In such a small sample, variability in both the quantity and the type of training participants were engaged in might produce spurious effects. Another aspect of the implementation of Omega-is-d2 training that was not systematically controlled in the present study was the amount of teacher support participants received. Teachers can elaborate on the comments from the user and on the content in Omega-is-d2, and by doing so help the user to construct a richer representational model of language content. In Rare Event Transactional Theory, such instances of recasting are assumed to increase the likelihood that learning will occur (Nelson, 1998). Further, teachers can also help resolve comprehension failures and discuss particularly difficult aspects of written language processing with the user. Based on models of language comprehension (Perfetti & Stafura, 2014; Rönnberg et al., 2013), such processes likely support developing language comprehension, and such discussions has been suggested to be crucial for DHH signing children who are learning to read (Hoffmeister & Caldwell-Harris, 2014; Svartholm, 2010). Thus, with more extensive training and better availability of teachers, effects on training might have been stronger in the present study. All these practical issues arose because we wanted to implement our intervention in a sign language rich learning environment, that is, the schools. Future studies should take steps to overcome practical hurdles in implementing reading interventions and determine the extent to which length of exposure to different types of exercises influences outcome.

Hierarchical linear modeling effectively deals with unequal spacing across test occasions, dependency in the data and power issues, and can include both time-varying and time-invariant predictors (Hesser, 2015; Singer & Willett, 2003). These features fitted well with the aim of the present study. However, spurious test–retest effects and missing data, especially at the first two test occasions, might have confounded the results on this small data set (Maas & Hox, 2005; McNeish, 2017). In other words, only tentative conclusions should be drawn from studies applying HLMs on small samples (Davis et al., 2013). A final limitation is the inclusion criteria for this study, since our selection of participants makes generalization from our sample to the general population of DHH signing children difficult. On the other hand, our participants represented 21% of the total population of DHH signing children at the schools invited to participate in this work, and we thus believe that our sample is representative of DHH signing children who are still struggling with learning to read in a Swedish context. The shortcomings addressed here can only be addressed by larger, less heterogeneous samples, and more complete data sets.

Conclusions

Although we did not find convincing evidence that our intervention supported reading development, a marginally significant effect on word reading was observed, which indicates that it is warranted to further investigate the use of Omega-is-d2 as a reading intervention for DHH signing children. Further, our data indicate that DHH signing children who are at an early stage of their reading development progress in both word reading and reading

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comprehension when followed over time. Active construction of novel lexical forms may be a mechanism working across modalities that underlies word reading development, and plausibly other processes comes into play in developing reading comprehension. Taken together, these results support the notion that reading skills, in particular word reading, are linked to sign language skills. Future intervention studies based on developing this link should ensure a firm base of sign language skill.

Supplementary Data

Supplementary material is available at *Journal of Deaf Studies and Deaf Education* online.

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Conflicts of Interest

No conflicts of interest were reported.

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