The relationship between visual speech perception, phonological awareness and reading in deaf and hearing children

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Declaration

I, Elizabeth Mary Worster, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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Abstract

Although some deaf children do achieve age-appropriate reading levels, on average deaf children's reading is poorer than that of their hearing peers. There are many factors that relate to reading ability in deaf children. The factor that is the focus of this thesis is speechreading (lipreading) skill. Perhaps surprisingly, speechreading ability also relates to reading ability in hearing children. The primary aim of this thesis is to investigate the relationship between speechreading and reading proficiency in both deaf and hearing children. The underlying working hypothesis is that information about the sublexical structure of speech can be extracted from visual speech and contribute to multimodal phonological representations, which can then support early single word reading.

The first study in this thesis used eyetracking to show that the time spent looking at the mouth during silent videos of speech correlates with deaf and hearing children's speechreading ability. The results suggest that deaf and hearing children access visual speech information in a similar way.

Subsequent studies used structural equation modelling to show that the relationship between speechreading and reading is mediated by phonological awareness in both deaf and hearing children. The final study adapted the Speechreading Training and Reading intervention games to show that speechreading can be trained in young hearing children. The children who performed poorest on a test of phonological awareness, phoneme blending, showed improvements on this task as a result of speechreading training.

This thesis furthers our understanding of the relationship between speechreading and reading in deaf and hearing children. This is of potential use to deaf and hearing children who have poor phonological skills and

therefore are likely to struggle with reading. These children may benefit from attention being drawn to visual speech information to improve their access to the sublexical structure of speech.

Impact statement

Reading is a fundamental skill that children are taught in the early stages of education. However, it has repeatedly been found that deaf children struggle to learn to read and that, on average, deaf children and adults have poorer reading skills than their hearing peers (Conrad, 1979; DiFrancesca, 1972; Qi & Mitchell, 2011; Wauters, Van Bon, & Tellings, 2006). Conrad (1979) found that deaf school leavers aged 16 had average reading levels of approximately 9 years old. Even though there have been recent advances in earlier diagnosis of children born deaf and in amplification devices, this does not appear to have been accompanied by a great improvement in phonological skills or reading ability for deaf primary school children (Harris, Terlektsi, & Kyle, 2017). This has clear implications for the general educational attainment of deaf individuals and their future employment options. Therefore, it is important to understand what factors relate to reading ability in deaf children. The factor that is the focus of this thesis is speechreading skill, which correlates with reading ability in both deaf and hearing children (e.g. Kyle, Campbell, & MacSweeney, 2016).

The work in this thesis furthers the understanding of the relationship between speechreading and reading and what the role of phonological awareness may be in this relationship. This has several implications for literacy development in both deaf and hearing children. The similarity in results between deaf and hearing children in each study described in this thesis is striking. The results suggest that deaf and hearing children access visual speech information in a similar way and that it also relates to reading ability in a similar way in these two groups.

For deaf children, the relationship between speechreading, phonological awareness and reading suggests that visual speech perception is important to consider. Therefore, practices that discourage a child's use of visual information during speech perception, such as discouraging them to pay attention to the face, are potentially restricting the child's ability to form robust multimodal phonological representations. This may have consequences for the development of their phonological awareness and reading skills. The studies in this thesis also suggest a potential role for visual speech perception in the development of reading in young hearing children. It is possible that emphasis on visual speech information may help hearing children with poorer phonological awareness to develop their phonological awareness skills by providing an additional source of information about the sublexical structure of speech.

Contents

List of figures

List of tables

1 Reading

1.1 Hearing children

The 'simple view of reading' (Gough & Tunmer, 1986) proposes that strong early language skills and decoding skills are needed to enable successful reading comprehension. If a child can identify and link the sounds represented in the text but does not have the vocabulary to understand what the word is they cannot access meaning from written text. Equally, if a child has a large vocabulary but is not able to decode the text they will not be able to understand the text. The importance of both of these aspects of reading is made clear through work with dyslexic children and poor comprehenders. Dyslexic children are characterised as children who have reading difficulties despite average or above average intelligence and good language skills, with a particular difficulty in decoding as opposed to comprehension (Snowling & Hulme, 1994; Snowling, 2000). Conversely, children with reading comprehension impairment show average decoding skills but below average language skills (for review, see Nation & Norbury, 2005). This demonstrates that both decoding and language skills are necessary for successful reading comprehension.

Models of adult skilled word recognition also support the idea that both phonological and language skills are important to reading proficiency (Castles, Rastle, & Nation, 2018). There are three key computational models of reading that have been proposed. The first is the Dual Route Cascade model, which is a model of skilled adult word reading (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001). The second is the Triangle model, which is a model of both the skilled adult reading system and the process of learning to read (Plaut, Seidenberg, & McClelland, 1996). The third model is the Connectionist Dual Process model (Perry, Ziegler, & Zorzi, 2007), which includes features of the previous two models. These models differ, but importantly all contain two key pathways for word reading: a direct route from orthography (text) to semantics (meaning) and an indirect phonological route that accesses semantics from orthography via phonology (sound), as shown in the Triangle model depicted in Figure 1. The three elements (orthography, semantics and phonology) in the model are thought to have bi-directional connections between them so that each can be used to access the other (Rastle, 2007).

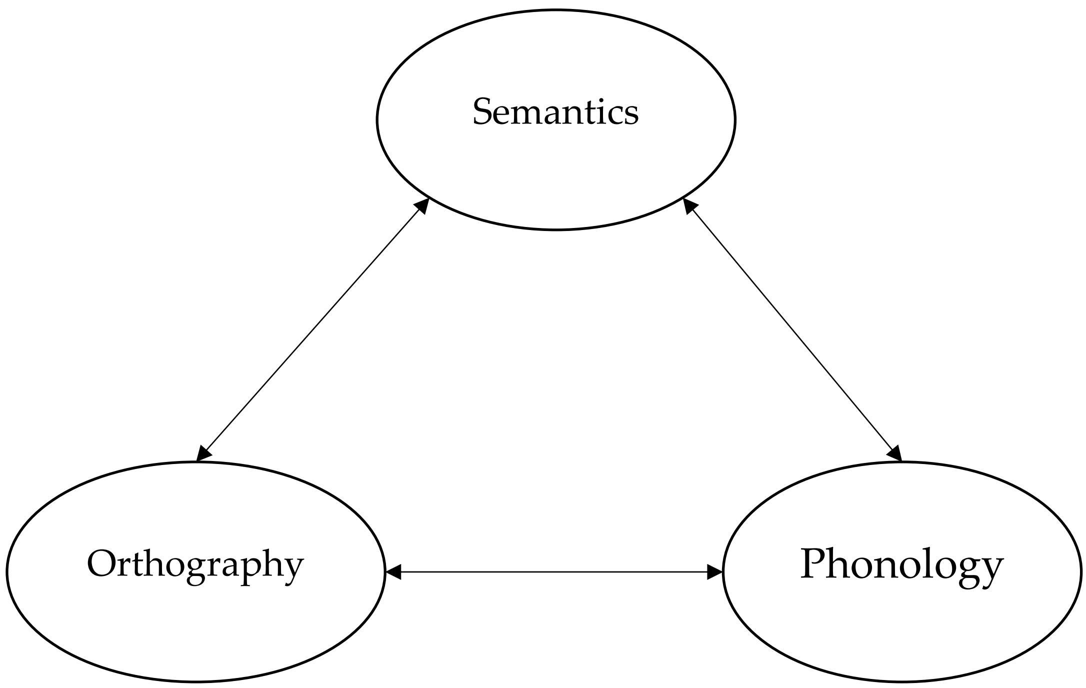

Figure 1. Schematic of the triangle model of reading.

In the Triangle model, when reading becomes automatic, the direct pathway seems to be preferred to access the meaning of words from text, rather than needing to go via the phonological route (Besner, Reynolds, & O'Malley, 2009). Equally, for irregular words, such as 'yacht', the word form may be mapped directly onto the meaning. However, when first learning to read, or as a fluent reader reading a new word, it is important to be able to map the text to sound in order to access the meaning. These models support the idea that both language skills, for semantic representations, and phonological skills, for the orthography-phonology route, are important for reading development.

In the following sections I will address the contributions of decoding and language skills to reading development and will describe the key sub-skills thought to underlie these two components of the simple view of reading.

1.1.1 Decoding

Hulme and Snowling (2013) summarised the literature on the early stages of reading development and concluded that there are three key factors that are important for the development of decoding: phonological awareness, letter-sound knowledge and Rapid Automatised Naming (RAN). These factors are highly correlated with one another and there is some debate as to whether they really measure different constructs (Torgesen, Wagner, Rashotte, Burgess, & Hecht, 1997; Wagner & Torgesen, 1987). However, many studies have shown that letter-sound knowledge, phonological awareness and RAN are independent predictors of reading development (Caravolas et al., 2012; Hulme, Bowyer-Crane, Carroll, Duff, & Snowling, 2012; Hulme & Snowling, 2013; Lervåg & Hulme, 2009; Norton & Wolf, 2012; Warmington & Hulme, 2012; Wolf, Bowers, & Biddle, 2000). Each of these three factors will be discussed in turn.

1.1.1.1 Letter-sound knowledge

The alphabetic principle refers to the idea that young children need to understand that individual letters and groups of letters represent particular sounds in spoken words (Byrne & Fielding-Barnsley, 1989). To learn to read alphabetic languages, such as English, it is important for children to master the alphabetic principle. In order to do this a child must be able to recognise letters and link them to sounds. Letter-sound knowledge can be considered to reflect paired-associate learning between an image on a page and a sound (Hulme & Snowling, 2013), allowing a child to sound out new words. Letter-sound knowledge has been shown to predict reading ability in typically developing children (Bond & Dykstra, 1967; Hulme, Bowyer-Crane, Carroll, Duff, & Snowling, 2012; Muter, Hulme, Snowling, & Taylor, 1998; Muter, Hulme, Snowling, & Stevenson, 2004). In addition, interventions that combine phonics training with a link to letter sounds tend to have larger effect sizes than those that use phonics alone (Ball & Blachman, 1991; Bradley & Bryant, 1983; Byrne & Fielding-Barnsley, 1989; Hatcher, Hulme, & Snowling, 2004). Letter-sound knowledge shows a particularly strong relationship with word recognition as opposed to reading comprehension (Muter et al., 2004).

1.1.1.2 Phonological awareness

Another key skill that underlies a child's acquisition of the alphabetic principle is phoneme awareness – the ability to isolate and manipulate phonemes in spoken words. This is especially the case in alphabetic languages, such as English (e.g. Goswami & Bryant, 1990; Melby-Lervåg, Lyster, & Hulme, 2012; Snowling, 2000) and Spanish, where to a first approximation letters in printed words map onto phonemes in spoken words (e.g. Hu, 2013). Additionally, the difficulties in reading that characterise developmental dyslexia are thought to arise primarily from a difficulty with phonological awareness (The phonological deficit hypothesis; Snowling & Hulme, 1994; Snowling, 2000). Phoneme awareness is one aspect of phonological awareness – how it is measured and how it relates to reading ability throughout development will be discussed in depth in Chapter 2.

1.1.1.3 Rapid Automatised Naming

Rapid Automatised Naming (RAN; Denckla & Rudel, 1976) is a speeded naming task which involves naming a set of items repeated randomly across a page. The items can be numbers, letters, colours or objects. It is a strong predictor of current and later reading ability in children (Norton & Wolf, 2012). Alphanumeric RAN (naming letters or numbers) is a stronger predictor of reading ability than non-alphanumeric RAN tasks (Compton, 2003). Although there are strong links between RAN and reading at many ages, there seem to be particularly strong links for early readers, around five to seven years old (Norton & Wolf, 2012). By this age most children have learnt the number and letter labels and thus can complete alphanumeric RAN tasks. RAN and letter-sound knowledge are closely related, which may be because they both tap an underlying cross-modal paired-associate learning mechanism (Hulme & Snowling, 2013). However, non-alphanumeric RAN with pictures or colours, measured before children learn to read, also predicts later reading ability (Lervåg & Hulme, 2009). Therefore, the relationship between RAN and reading cannot be attributed solely to letter-sound knowledge. RAN is a particularly strong predictor of children's reading fluency, but also predicts reading accuracy and measures of fluency that require reading comprehension (Norton & Wolf, 2012; Schatschneider, Fletcher, Francis, Carlson, & Foorman, 2004). This suggests that the relationship between RAN and reading cannot be explained only by a child's general speed of processing (Hulme & Snowling, 2013). Instead, RAN is thought to tap the retrieval of phonological representations from long-term memory suggesting that perhaps it should not be differentiated from phonological awareness in predicting reading ability (Wagner & Torgesen, 1987; Wimmer, Mayringer, & Landerl, 2000). However, as described above, although RAN and phonological awareness are closely related, they are independent predictors of early reading ability.

1.1.2 Language skills

Decoding printed words into speech (even inner speech) is an important skill that is perhaps the first step for hearing children learning to read. However, if a child does not have sufficient language skills in order to understand what they have decoded they are limited in how much meaning they can access through written text.

The size of a child's vocabulary, both receptive and expressive, is particularly important for learning to read, showing a relationship with concurrent reading ability and also predicting later literacy outcomes, even from early measures of vocabulary (Lee, 2011; Lervåg & Aukrust, 2010). Although children who have better vocabulary skills tend to be better at decoding (Verhoeven, van Leeuwe, & Vermeer, 2011), the relationship between vocabulary size and reading comprehension is stronger (Muter et al., 2004). Training studies have suggested that the relationship between language ability and reading is causal, showing that training in oral language skills leads to improvements in reading comprehension (Fricke, Bowyer-Crane, Haley, Hulme, & Snowling, 2013). As well as allowing a child to understand the words that they have decoded, Wegener et al. (2018)

suggested that oral vocabulary knowledge combined with a child's letter-sound knowledge can make decoding easier as the child can predict the form of the word based on their knowledge of how it sounds. Wegener et al. (2018) orally trained children on a set of novel non-words and then asked them to read sentences containing those words and other words. They showed that when the children read the words for the first time they looked at the trained words for less time than untrained words, but only if they had regular spelling. This supports the idea that oral vocabulary aids both decoding and comprehension of written words by allowing the child to form predictions about word forms.

Duff and Hulme (2012) conducted two studies to investigate the effect of phonology and semantics on learning to read words. In their first study they found that a child's semantic knowledge of a word predicted how well they would learn to read it. However, they then pre-trained 5-to-6-year-old children with phonological information and semantic information. They found that teaching the sounds of the words aided learning to read that word out loud but that pre-training the meaning of the word did not add any additional benefit in written word learning. Although this study might suggest semantic knowledge has limited impact on word recognition, vocabulary is very important for comprehension. Indeed, Monaghan and Woollams (2016) noted that phonological training can only transfer to reading comprehension if the reader already has good oral language. They tested a connectionist triangle model of reading to investigate how damage to either the semantic representations or phonological representations would affect the model's performance. Monaghan and Woollams (2016) found that impairments in any part of the model resulted in reading difficulties throughout the model, so poor vocabulary resulted in deficits in phonological processing and vice versa. They suggested that this was because the model pre-training only allowed mapping between phonology and semantics, simulating vocabulary learning in pre-readers, thus strengthening the pathway between them. Monaghan and Woollams (2016) suggested that pure deficits in reading (either in decoding or comprehension) are unlikely to exist as phonological and semantic representations interact with each other.

The relationship between vocabulary and reading ability is likely to be causal in both directions. A bigger vocabulary may help a child to make predictions about the text and therefore aid decoding as well as their comprehension. In turn, reading will expose a child to a larger range of words and thus improve their spoken vocabulary (Verhoeven et al., 2011). This can lead to a Matthew effect (Stanovich, 1986). This refers to a widening in the gap in reading ability between better and poorer readers, as children entering school with a lower vocabulary are likely to struggle more to learn to read and in turn will learn fewer words, resulting in the gap widening as children progress through school (Nation, 2017).

Although language skills, in particular vocabulary, are necessary for learning to read, the studies in this thesis focus on single word reading accuracy. Therefore discussions in this thesis will focus on decoding rather than on comprehension. Specifically, I will focus on the role that phonological awareness plays in the development of single word reading in both deaf and hearing children.

1.2 Deaf children

In this section I will give a brief overview of issues relating to deafness and will discuss the impact deafness can have on reading ability. I will discuss what technological advances in both screening and amplification technology mean for reading development in children born deaf. Finally, I will address each of the factors outlined above that are important for hearing children to learn to read and discuss how their role might be similar or different in deaf children's reading development.

1.2.1 Types of deafness

Deafness can vary on different dimensions, including the age of onset of deafness, the severity of deafness, the permanence of deafness, and its aetiology. Hearing level is measured in decibels (db), indicating the quietest sound that a person can hear. Deafness is categorised into four levels of severity: mild (25-39db), moderate (40-69db), severe (70-94db) and profound (95+db). Most conversational sounds occur within the 45-60db range, so children with moderate hearing loss may have some difficulty with hearing speech and children with severe-to-profound hearing loss will have substantially reduced access to speech sounds. This thesis focuses on children with a permanent moderate-to-profound hearing loss with an onset before the age of 12 months.

1.2.2 Reading achievement in deaf children

1.2.2.1 Early studies

Deaf children and adults have been shown to have poorer reading skills, on average, than their hearing peers (Conrad, 1979; DiFrancesca, 1972; Qi & Mitchell, 2011; Wauters et al., 2006). A nationwide study from nearly 40 years ago (Conrad, 1979) found that deaf school leavers aged 16 had average reading levels of approximately 9 years old. Since then, studies have indicated that the gap in attainment between deaf and hearing children has remained large despite adjustments to measurements and advances in technology (Harris et al., 2017a).

Qi and Mitchell (2011) reviewed deaf and hard-of-hearing students' maths and reading performance on the Stanford Achievement Test from 5 timepoints over 30 years (1974-2003). They found that although the test was adapted for use with deaf and hard-of-hearing students, the deaf children aged 8-18 years consistently performed worse than their hearing peers on both reading and maths but that the gap in attainment was larger for reading than for maths. In addition, 18-year-olds had average reading levels of a grade 4 standard (9 years old), demonstrating the severity of the gap. This suggests that on standardised measures of reading, the gap between deaf and hearing children's performance has not closed over time. However, this may be partly due to the measurement process, as standardised measures of reading and reading-related skills often require children to provide a verbal output. This can lead to underestimation of their reading skills as it is confounded with their speech output ability. Although deaf children showed poorer reading skills on average than their hearing peers, there is substantial variability in reading skill within this group, with some deaf children showing similar reading ability to their hearing peers (Kyle & Harris, 2010).

Since 2003, the last timepoint in Qi and Mitchell's (2011) study, there have been two key areas of advancement: the age of diagnosis of deafness and the type and quality of amplification.

1.2.2.2 Age of diagnosis

The universal newborn hearing screening programme (UNHS) was introduced in the United Kingdom in 2006. Early assessment of district-level newborn hearing screening prior to a national rollout indicated that newborn hearing screening reduced the age at which children were identified as having hearing loss from an average of 17 months to just a few weeks old (Davis et al., 1997). This hearing screening is considered to be highly effective at identifying newborn hearing loss when a confirmatory follow-up check is carried out (from 31% identified before 6 months without newborn hearing screening to 74% identified before 6 months with newborn hearing screening; Kennedy, McCann, Campbell, Kimm, & Thornton, 2005). This means that, whereas previously children may have missed out on a year and a half of language input before their deafness was identified, parents now know very early whether their child is deaf and can start to consider options for supporting their child's language development at a much earlier age.

The Wessex trial (Kennedy et al., 2006) measured language ability of children involved in newborn hearing screening trials and those who had not. They found that deaf children who were identified before the age of 9 months had better language skills than those who were identified later and that involvement in the hearing screening trial was associated with better receptive language outcomes. These findings have been supported in reviews of hearing screening in both America and Australia (Pimperton & Kennedy, 2012). Improved language skills are likely to impact literacy skills and early identification of deafness means children can also receive aiding at an earlier age, which in turn can impact literacy attainment as discussed below. The deaf children involved in this thesis had an average age of 9 months old when they were identified as deaf.

1.2.2.3 Amplification

Along with earlier diagnosis, there have been big changes in recent years in the technology used for amplification.

In the UK, since the late 1990s any child eligible for cochlear implants (CIs) can receive them unilaterally on the NHS. A CI works by directly stimulating the auditory nerve with up to 22 stimulating electrodes. This number is minimal compared to the 16,000 hair cells in a typical cochlea, meaning that although access to auditory speech information is greatly improved the auditory access children receive through CIs is still limited. 74% of eligible children in the UK now receive implants before the age of 3 and 94% by the age of 17 (Raine, 2013).

Some studies have demonstrated that having a CI benefits language skills, such as vocabulary and syntax, as well as speech and hearing in deaf children (Cupples et al., 2017; Geers & Nicholas, 2014). Furthermore, earlier implantation is associated with better language outcomes (Geers & Nicholas, 2014). However, in terms of literacy outcomes the picture is less clear (Harris, 2016). Some deaf children with CIs appear to develop reading at the same rate as their hearing peers (Archbold et al., 2008; Geers, 2003), although this is not necessarily maintained into adolescence (Geers, Tobey, Moog, & Brenner, 2008; Harris & Terlektsi, 2011).

Hearing aids are also still commonly used to amplify sounds for children born deaf. Hearing aids amplify the sound reaching the ear and are therefore only suitable for children who have an intact auditory pathway. Again, evidence for an influence on literacy outcomes is mixed with some studies suggesting that children with CIs show better literacy skills than their peers with hearing aids (Thoutenhoofd, 2006; Vermeulen, van Bon, Schreuder, Knoors, & Snik, 2007) and others showing that with the introduction of digital hearing aids (Ackley & Decker, 2006) children with hearing aids show better reading skills than those with CIs (Harris & Terlektsi, 2011).

In summary, in the last two decades there have been some key advances in identification of deaf children and the amplification devices available. Early identification improves language outcomes for deaf children, which is likely to influence literacy attainment. Advances in cochlear implants and hearing aid technology has increased deaf children's access to auditory speech to some extent and also improves language outcomes. However, the influence of aiding on literacy attainment is less clear. In the studies in this thesis, approximately half the deaf children had cochlear implants and half had hearing aids, with just a few with no aiding at all.

1.2.2.4 Recent studies

Despite advances in recent years in hearing screening and aiding for children born deaf, this does not yet seem to have affected average phonological awareness and reading development in deaf children and they still show poorer average reading skills than their hearing peers (e.g. Kyle & Harris, 2010; Kyle, Campbell, & MacSweeney, 2016; Lederberg, Schick, & Spencer, 2013). Harris et al. (2017a) studied two cohorts of severely-to-profoundly deaf children ten years apart and a comparison group of hearing children. The deaf children were 5-7 years old and the hearing children were matched to the group based on single word reading scores. The children completed measures of reading, phonological awareness and vocabulary. Harris et al.

(2017a) found that the later cohort had higher vocabulary scores than the deaf children tested ten years previously but there were no differences in either phonological awareness or reading. Both groups of deaf children also had lower scores than their hearing peers on both of these measures. This suggests that despite the advances in technology to aid deaf children in recent years, deaf children will continue to need additional support to develop their reading.

1.2.2.5 Summary

In summary, early studies indicated that on average deaf children have poorer reading skills than their hearing peers. Despite earlier identification of deafness and advances in technological aiding, more recent studies suggest that this gap in reading attainment still exists between deaf and hearing children. This has clear implications for the general educational attainment of deaf individuals. However, there remains huge variability in the reading ability of deaf children and adults. There are several factors that account for some of this variability in deaf children's reading ability, including severity of deafness, age of onset, age of diagnosis and aiding and language used at home and in school.

1.2.3 Predictors of reading ability in deaf children

Given all of the additional factors that can contribute to a deaf child's reading attainment, it is important to understand whether the same skills required for hearing children to read are also important for deaf learners.

1.2.3.1 Phonological awareness

The role of phonological awareness in deaf children's reading development will be discussed in depth in Chapter 2. There is extensive debate over whether deaf children use phonology when reading (e.g. Mayberry, del Giudice, & Lieberman, 2011; Mayer & Trezek, 2014). As with hearing children, performance on phonological awareness tasks does predict reading ability in deaf children, accounting for approximately 11% of the variance in reading scores (Mayberry et al., 2011; Mayer & Trezek, 2014). Despite this relationship between performance on phonological awareness tasks and reading ability it is still debated whether deaf individuals automatically access phonology during reading (Bélanger, Baum, & Mayberry, 2012; Chamberlain, 2002; Hanson & Fowler, 1987; Mayberry et al., 2011).

1.2.3.2 Letter-sound knowledge

Deaf children tend to have poorer letter-sound knowledge than their hearing peers. However, as with hearing children, letter-sound knowledge is a predictor of reading ability in deaf children both for concurrent reading ability (Easterbrooks, Lederberg, Miller, Bergeron, & Connor, 2008; $r = .63$) and as a longitudinal predictor (Kyle & Harris, 2011; $r = .48$). For deaf children, letter-sound knowledge may provide the building blocks for phonological awareness. The relationship between orthography and phonology in deaf children will be discussed in Chapter 2.

1.2.3.3 *Rapid Automatised Naming*

Relatively little is known about the role of RAN in deaf children's reading ability. The only published study in this area found that RAN skills in speech or sign did not relate to deaf adolescents' reading ability despite relating to reading in reading-age-matched hearing controls (Dyer, MacSweeney, Szczerbinski, Green, & Campbell, 2003).

1.2.3.4 *Vocabulary/language ability*

Language skills are important for all children learning to read, and perhaps even to a greater extent for deaf children than for hearing children given their relatively poor phonological skills. Although there is large variability in hearing children's language ability when they start school, the language background of deaf children shows much more extreme variation.

As discussed above, many deaf children will receive a cochlear implant or hearing aid and will use spoken communication both at home and at school (The Ear Fountation Report to Oticon; Allen, Yen Ng, & Archbold, 2016). However, in order to access spoken language, deaf children rely on any aided hearing level they may have and on speechreading, both of which are limited sources of information. Although many deaf children do develop good spoken communication, many others do not, meaning that within the population of deaf children who learn a spoken language there is huge variability in their spoken language outcomes (Harris, Terlektsi, & Kyle, 2017b; Tomblin et al., 2015).

In the UK, British Sign Language (BSL) is the preferred language of approximately 70,000 Deaf people. The capital 'D' for Deaf here refers to cultural Deafness, which includes the use of BSL and belonging to the Deaf community. As the children in this thesis were from a mixture of backgrounds the lower case 'd' will be used from here on to refer to that group because the group is defined by hearing status and not by culture. Signed languages are natural human languages that are different in different countries and are not based on the host spoken language. BSL was officially recognised by the British government as a language in 2003 and expresses linguistic content through a mixture of hand shapes, facial expressions and movement of the upper body.

Approximately 90% of deaf children are born to hearing parents (Mitchell & Karchmer, 2004), who most likely do not have any knowledge of a signed language. Deaf parents of a deaf child are likely to communicate with their child using a signed language. In this case, the child has full access to a rich language environment and can develop language naturally, hitting the same developmental milestones as a hearing child learning a spoken language (Emmorey, 2002). In addition, children who use BSL will be bilingual to some extent as spoken English is the dominant language in the surrounding community and they must learn to read a spoken language.

As well as English and BSL, some parents will use a combination of the two in communication methods such as Sign-supported English (SSE), which uses lexical signs from BSL while using spoken English as the matrix language. Some parents will use spoken English with Cued Speech (Cornett, 1967), which involves a set of manual gestures that represent different phonemes. This combination of speechreading and hand movements gives a deaf child full access to the phonological structure of spoken English.

The decisions parents make about how to communicate with their deaf child will depend on several factors such as the child's hearing level, the parents' hearing status and their knowledge of a sign language and the Deaf community, as well as what services they have locally to support them and their child. This leads to deaf children being a very heterogeneous group in terms of their language background and language proficiency. On average though deaf children, who do not have a signed language as a native language, show severe language delays (Kyle & Harris, 2006), which are likely to be a key contributor to their reading difficulties.

1.2.3.5 The role of language in deaf children's reading acquisition

Vocabulary measures of both sign and speech are a strong predictor of reading outcomes for deaf children both concurrently (Easterbrooks et al., 2008; Harris & Beech, 1998; Kyle et al., 2016; Kyle & Harris, 2006) and longitudinally (Kyle & Harris, 2010, 2011). Mayberry et al. (2011) suggested that language ability, indexed by a combination of measures including spoken and sign language vocabulary production and also comprehension, is the strongest predictor of reading ability in deaf children, accounting for 35% of variance in reading skill.

Some studies report that sign language ability is positively correlated with reading achievement (Goldin-Meadow & Mayberry, 2001; Hermans, Knoors, Ormel, & Verhoeven, 2008; Strong & Prinz, 1997). Others report that better spoken language proficiency relates to better reading (Kyle & Harris, 2006, 2010, 2011). These results suggest that strong language skills, regardless of the modality, are important for successful reading development (e.g. Kyle, 2015).

Geers et al. (2017) examined language outcomes in deaf children with CI. The authors compared reading ability in orally educated deaf children to that in children exposed to sign language and found that the orally educated deaf children had better reading skills. However, in this study the group of children exposed to sign language included any child who had been exposed to some form of manual communication, including baby signs, home signs and signed English. This group of children exposed to sign language are likely to have dramatically different language experiences from one another in terms of the onset, duration and quality of their sign language exposure. They are therefore not comparable to native signers, who have a strong first language. Treating native signers and children exposed to some signs as a single group confounds the relationship between sign language and reading with the relationship between language proficiency and reading.

Good language skills are clearly important for learning to read. However, although having a strong foundation in any first language is likely to benefit a deaf child learning to read, the written word is a visual representation of spoken language. Therefore, even if a child has strong sign language skills they will still have to develop some understanding of the spoken language in order to develop good reading skills (Hermans, Ormel, & Knoors, 2010; Lederberg et al., 2013).

1.3 Chapter summary

In summary, in order for children to learn to read it is important for them to be able to both decode printed words and have the language skills to comprehend what they can decode. Deaf children on average show a deficit in reading skills compared to their hearing peers that increases with age.

Deaf children are an extremely heterogeneous group on several levels including their age of diagnosis, residual hearing level, language background and proficiency and what type of aiding they have, all of which can affect their reading ability. There are several factors that explain the variation in hearing children's reading, including phonological awareness, letter-sound knowledge and vocabulary skills. The role of each of these factors in reading development is fairly well established for hearing children. However, the extent to which each of these factors contributes to reading ability in deaf children is less clear. For example, although important, phonological awareness is likely to make less of a contribution to early reading development in deaf children than in hearing children. This is because they have reduced access to spoken language in order to develop robust phonological representations. In the following chapter the role of phonological awareness in hearing children's reading development will be reviewed before considering whether phonological awareness is also important for deaf children's reading development.

2 Phonological awareness

The previous chapter outlined several key factors that relate to reading development in both deaf and hearing children. As the relationship between phonological awareness and reading is a key focus of this thesis, this chapter will look in greater depth at phonological awareness as a factor related to reading development. Phonological awareness is the ability to represent and manipulate the sounds in spoken words. This awareness enables these speech sounds to then be mapped, in alphabetic languages, to the graphemes on the page, thus allowing decoding of the printed word. This chapter will address how phonological awareness skills may be subdivided, how they are measured and different ways in which they may relate to reading ability in hearing children. It will then address the debate as to whether and how it might relate to reading development in deaf children. Finally, I will discuss the multimodal nature of phonological representations. There is a substantial amount of work investigating whether skilled readers automatically access phonological information when reading words. This is a particularly debated topic for deaf skilled readers, with much research suggesting that they do not access phonological information. This topic is beyond the scope of the body of this thesis.

2.1 Hearing children

In many models of literacy development (e.g. Ehri, 1995; Frith, 1985; Jackson & Coltheart, 2001) there are three key stages a child goes through when learning to read. According to Frith (1985) there is an initial logographic stage, where they recognise simple or common words as wholes, and then an alphabetic stage involving phonic decoding, where they learn that letters represent sounds and so learn to break down words. Finally, skilled readers reach an orthographic stage, where they are able to automatically access the meaning of words from print without having to sound them out.

Phonological skills are strongly related to reading ability in hearing children. This has been shown through many concurrent, longitudinal and intervention studies (for a detailed review, see Melby-Lervåg, Lyster, & Hulme, 2012) and phonological difficulties are a core symptom of developmental dyslexia (Snowling, 2000). A full review of phonological awareness and developmental dyslexia is beyond the scope of this thesis, but there are several key points to highlight about how phonological awareness is measured and in what way it relates to reading ability in hearing children.

First, it is important to distinguish between explicit phonological awareness and implicit phonological coding. Explicit phonological awareness refers to the ability to represent and manipulate sound units within words. Implicit phonological coding refers just to the representation of sound units, but not the ability to reflect on them and manipulate them (Melby-Lervåg et al., 2012). Implicit phonological coding is measured using verbal short-term memory tasks (e.g. digit span) and non-word repetition tasks, which all require access to phonological representations. Early performance on these implicit phonological coding tasks predicts later development of word reading skills (Gathercole & Baddeley, 1993; Hulme & Snowling, 2013) and interacts with reading development. Non-word repetition improves with learning to read (Nation & Hulme, 2011). For example, literate adults have better non-word repetition performance than illiterate adults (Reis & Castro-

Caldas, 1997). In this thesis, only explicit phonological awareness was measured, but the relationship with underlying phonological representations will be discussed in the experimental chapters.

Explicit phonological awareness tasks are thought to tap into the same underlying phonological representations as implicit phonological coding tasks. Explicit phonological awareness tasks have been used to assess children's phonological skills including segmenting and blending and vary in the level of phonological unit they assess. Words can be broken down on the basis of syllables, onset and rime units and by individuals phonemes. The onset of a word refers to the initial consonant sound or cluster and the rime unit includes the following vowel and consonants. For example, the word 'sheep' can be broken down into 'sh' (onset) and 'eep' (rime). Larger units (syllables or rime) are thought to be easier for children to manipulate than smaller units (phonemes) (e.g. McBride-Chang, 2004). Most studies in the literature have assessed rime-level or phoneme awareness, therefore the role of syllable awareness will not be discussed further here. Often both rime-level awareness and individual phoneme-level awareness, also known as phonemic awareness, are assessed and are referred to together under the umbrella term of phonological awareness. However, several researchers have suggested that phonological awareness can be subdivided and Melby-Lervåg et al. (2012) outlined three competing theories about the relationship between different levels of phonological awareness and reading development. The first theory is that rime-level awareness is important for the first stages of reading development and that phonemic-level awareness only develops as a result of learning to read (Goswami & Bryant, 1990). The second theory suggests that phonemic-level awareness is important for learning to read and that it continues to develop as a result of learning to read (Hulme, Caravolas, Málková, & Brigstocke, 2005). The third theory suggests that phonological awareness can be considered as a single factor that predicts reading skill, without making distinctions between rime-level awareness, phonemic-level awareness and syllable awareness (Anthony et al., 2002; Anthony & Francis, 2005; Anthony & Lonigan, 2004; Papadoupoulos, Spanoudis, & Kendeou, 2009). However, Melby-Lervåg et al. (2012) found in their meta-analyses that phonemic-level awareness was the strongest correlate of reading ability in children, over rime-level awareness and also verbal short-term memory skills. This suggests that phonological awareness is not just one factor but can be divided into different sub-components. In addition, Melby-Lervåg et al. (2012) reported that phonemic awareness and rime-level awareness are both longitudinal predictors of word-reading ability in pre-school children (Melby-Lervåg et al., 2012; National Institute for Literacy, 2008), with phonemic awareness being a stronger predictor (r = .43, r = .42 respectively) than rime-level awareness (r = .28, r = .29 respectively). Therefore, it seems that early phoneme-level awareness and early rime-level awareness are related to reading ability, although the relationship is stronger for phoneme-level awareness.

The studies described above support the idea that early phonological skills do predict later reading development (Melby-Lervåg et al., 2012). There is also substantial evidence that phonological representations and skills develop as a result of learning to read (e.g. Castles & Coltheart, 2004; Nation & Hulme, 2011). However, these two ideas are not mutually exclusive, as phonological skills may have a reciprocal relationship with reading, with early phonological skills predicting reading development and the further development of phonological awareness then being facilitated as a result of learning to read. Hogan, Catts and Little (2005) found that phonological awareness in kindergarten (5-6 years old) predicted word reading skills in second grade (7-8 years old) but that phonological awareness in second grade did not predict word reading in fourth grade (9-10 years old) when controlling for word reading in second grade. In addition, second grade word-reading skills predicted fourth grade phonological awareness skills. This supports the idea that there is a reciprocal relationship between reading and phonological awareness (Perfetti, Beck, Bell, & Hughes, 1987).

2.1.1 Impact of research on phonics instruction

As summarised above, over the past few decades there has been growing evidence that phonological processing is important for reading development. In 2006, a review of reading and dyslexia commissioned by the UK government recommended that 'synthetic phonics' be used in schools (Rose, 2006). 'Synthetic phonics' involves teaching children to sound out words on a letter-by-letter basis and then blend those sounds together. This approach to teaching is now implemented in schools across England. During the process of training teachers to deliver the 'synthetic phonics' programme, Machin, McNally and Viarengo (2018) evaluated the effect of this phonics intervention. As the rollout of the intervention was staggered across the country they were able to compare reading achievement (from teacher assessment and national curriculum levels) in children educated with 'synthetic phonics' and those educated with previous teaching practices. They found that the 'synthetic phonics' group showed improved reading skills compared to the control group at ages 5 and 7 (treatment effect $\beta = 0.3$ standard deviations and 0.07 standard deviations respectively). At age 11 there was no group-level effect of the intervention. However, the treatment effect was still evident at age 11 for children who were at higher risk of reading difficulties and therefore reduced the overall gap in reading attainment. All children in England now have to take the Phonics Screening check at the end of Year 1 (6-7 years old) in which they have to read words and pseudowords to assess their decoding skills. As a result, teaching phonics has been made a priority in the first years of primary school.

2.2 Deaf children

Given the strong focus on phonological awareness skills in hearing children in the last few decades, this has also been a focus of reading research with deaf children. For deaf children there is extensive debate about how they perform on phonological awareness tasks and how this relates to reading development. As they have reduced access to auditory information, it is likely that many deaf children will have poor representations of the phonological patterns of a spoken language and therefore may learn to read through a different route, for example by recognising whole words rather than by sounding them out. However, having reduced access to auditory information does not automatically exclude a child from developing phonological awareness. It is possible that phonological representations are multimodal, drawing on several sources of information (discussed later in Section 2.3). In the following section I will discuss deaf children's performance on phonological awareness tasks and how this may relate to reading.

Unsurprisingly, in general deaf people show poorer performance on phonological awareness tasks than their hearing peers. For example, Sterne and Goswami, (2000) showed that profoundly deaf adolescents had poorer phoneme-level awareness than reading-age-matched hearing controls (62%

vs. 92%) as measured by a pseudohomophone-to-picture-matching task. Despite this, the deaf adolescents performed above chance level on syllable, rhyme and phoneme tasks. Several studies have indicated that younger deaf children show evidence of phonological awareness in explicit phonological awareness tasks, such as rhyme-judgement and pseudohomophone-to-picture-matching tasks (Dyer et al., 2003; Harris & Beech, 1998; Kyle & Harris, 2006). However, even though the deaf children in these studies showed evidence of phonological awareness, they typically showed poorer performance than their hearing peers on these phonological awareness tasks. Other studies have indicated that phonological awareness skills vary with the level of segmentation required, with larger segments, such as syllables, being easier than smaller segments, such as phonemes. For example, James, Rajput, Brinton and Goswami (2008) found that 7-year-old deaf children who had received a cochlear implant when they were between 2 and 3 years old showed similar syllable and rhyme judgement to reading-age-matched hearing children. However, they showed poorer performance than the hearing controls on phoneme-level judgements, supporting the idea that larger segments may be easier for deaf children to show awareness of than individual phonemes. In addition, James et al. (2008) found that 9-year-old deaf children who received a cochlear implant when they were between 5 and 7 years showed similar syllable judgement to reading-age-matched hearing children but showed poorer performance on both rhyme and phoneme judgement tasks. Given that the later implanted group were also older than the early implanted group, it is difficult to draw conclusions about the effect of age of implant. However, overall, syllable judgement seems to be similar in deaf children to reading-age-matched hearing children. In contrast, rhyme and phoneme judgement is generally poorer compared to reading-age-matched controls. The same was true in comparison to chronological-age-matched controls. Similar results were found by Johnson & Goswami (2010), who tested 5-to-15-year-old deaf children with cochlear implants on rhyme and phoneme judgement tasks. They found that approximately half of the children with cochlear implants performed above chance level on the rhyme-judgement task. In addition, they found that children who received their cochlear implant before the age of 3 showed equivalent rhyming skills to reading-age-matched hearing children. However, there was no difference in performance between the children implanted before the age of 3 and those implanted later. Although, the early implanted children showed similar performance to reading-age-matched hearing children on the rhyme-judgement task the deaf children showed poorer performance on initial phoneme (onset) judgement regardless of when they received a cochlear implant. This suggests that rhyme awareness may be easier to establish for the deaf children than phoneme-level awareness.

One factor that is thought to improve phonological awareness in deaf children is the use of Cued Speech (see section 1.2.3.4), as the combination of mouth movements and hand gestures provides the child with full access to the phonetic patterns in speech. Charlier & Leybaert (2000) found that 10-to-13year-old deaf children who used Cued Speech at home and in school showed equivalent rhyme-judgement skills to hearing controls in the same year group. In addition, they showed better performance on the rhyme-judgement task than children who used Cued Speech only in school and those who did not use Cued speech at all. This suggests that early and consistent access to the speech patterns supports the development of phonological awareness.

So far in this chapter phonological awareness tasks have been discussed in relation to the level of word segmentation they involve. Phonological awareness tasks also vary in the type of manipulation they require. For example, common phonological awareness tasks include blending (putting presented phonemes or syllables together into a word), segmenting (breaking down a word into its phonemes or syllables), isolation (identifying a sound in a particular position in a word) and deletion (removing particular sounds from a word and repeating it). Carroll & Breadmore (2017) found that children with otitis media, who have a history of repeated ear infections and often have some difficulty with literacy skills, showed a specific deficit in segmenting and blending tasks but not in phoneme manipulation tasks such as deletion. This suggests that different manipulations require different skills and therefore may relate to reading ability in different ways. As explicit phonological tasks require some form of manipulation of sounds they will require additional skills beyond accessing phonological information, and these may vary across tasks. Therefore, it is important to recognise how phonological awareness is measured in different studies and how that may affect the results.

There are some limitations in assessing phonological awareness in deaf children. Typically, standardised assessments of phonological awareness are reliant on auditory access and verbal output. For example, in sound deletion tasks a child might be asked 'Can you say bat without the /b/' requiring them to hear the instructions and say the word. However, this style of task is not appropriate for use with all deaf children as it confounds their phonological awareness with both their ability to hear the experimenter and their speech production ability. In the studies mentioned above, phonological awareness was typically measured with picture-matching tasks, which avoid this difficulty. However, picture-matching tasks are typically multiple-choice and therefore open up the possibility of children just guessing the answer making them less sensitive as a measure. In addition, as different studies tend to devise their own tasks, it is hard to make comparisons across studies about phonological awareness performance.

The studies described above suggest that deaf children do show evidence of phonological awareness, albeit to a lesser extent than hearing children. However, there is still conflicting evidence about whether and how this relates to reading ability. Some studies have found a moderate positive correlation between performance on phonological awareness tasks and reading ability (r = .54, Campbell & Wright, 1988; Conrad, 1979; rhyme judgement r = .39, pseudohomophones r = .46, Dyer et al., 2003; r = .43, Harris & Beech, 1998). However, several other studies have not found a significant relationship between phonological awareness and reading ability in deaf children (e.g. Hanson & Fowler, 1987; Kyle & Harris, 2006; Leybaert & Alegria, 2003; Mayberry et al., 2011). Mayberry et al. (2011) conducted a meta-analysis of studies assessing the relationship between reading ability and phonological coding and awareness skills in deaf children. They included a range of phonological tasks, such as rhyme- and pseudohomophone-judgement and lexical decision tasks. They found that approximately half the studies found evidence for a relationship between phonological coding and awareness and reading and half the studies did not. However, the average effect size across the studies was z = 0.35 (where z is a logarithmic transformation of r values across the studies) with phonological coding and awareness skills explaining 11% of the variance in children's reading scores. Mayberry et al. (2011) concluded that phonological coding and awareness skills are related to reading ability in deaf children, but that they only explain a small amount of variance. However, Bus and Van Ijzendoorn (1999) conducted a meta-analysis of studies of hearing children and found that in hearing children a similar amount of variance (12%) in word-reading scores was accounted for by phonological awareness skills. Mayer and Trezek (2014) compared these two meta-analyses (Bus & Van Ijzendoorn, 1999; Mayberry et al., 2011) and noted that despite similar amounts of variance in reading scores being explained in both deaf and hearing children these meta-analyses reached opposing conclusions. Mayer and Trezek (2014) reviewed several studies regarding the relationship between phonological coding and awareness and reading ability and concluded that phonological skills play a similar role in reading development in deaf and hearing children. They also highlighted that the meta-analysis by Mayberry et al. (2011) involved studies across a wide age range, from children to adults, but that phonological skills are known to be of particular importance for beginner readers. Although there is some evidence that phonological awareness relates to reading in deaf children, this is based on concurrent correlational studies and therefore the direction of this relationship is unclear. Longitudinal studies can help to clarify the likely direction of effects.

Several studies have suggested that deaf children develop their phonological skills as a result of learning to read, rather than the other way around. Kyle & Harris (2010) conducted a 3-year longitudinal study with severely-to-profoundly deaf 7-8-year-olds to investigate the relationship between reading, speechreading, vocabulary and phonological awareness. They found that earlier reading ability predicted later performance on alliteration- and rhyme-judgement tasks, but early performance on these phonological awareness tasks did not predict later reading ability. They suggested that deaf children may use orthographic information to develop their phonological awareness. Indeed, when making judgements about phonological similarity deaf individuals show a high reliance on orthographic information. For example, Campbell & Wright (1988) presented deaf adolescents with a rhyme-judgement task with written words that had congruent spelling (e.g. bat/hat) and incongruent spelling (e.g. hair/bear). They found that overall the deaf adolescents had a particular difficulty identifying rhyming words that were spelled incongruently. The authors suggested that orthographic information may provide deaf individuals with important additional information about the patterns in spoken language beyond what they can gain from speechreading and residual hearing. This supports the idea that learning to read may improve phonological representations in deaf individuals. As discussed with hearing children, it is likely that the relationship between reading and phonological awareness is reciprocal in deaf children, with early phonological skills predicting reading ability and learning to read improving phonological awareness. It is likely that deaf children rely on orthography to a greater extent than hearing children to develop their phonological awareness skills as orthography provides a visual representation that may aid the development of an awareness of the sublexical structure of the spoken language. This will be discussed in the following section.

2.3 Multimodal phonological representations

So far phonological representations have been discussed in terms of sound patterns. Auditory information is undoubtedly the most useful information when it comes to understanding the sublexical structure of spoken words. However, there are also other sources of sublexical information, including visual speech, orthography and articulation. These different sources of information interact with each other to achieve speech perception (see McGurk effect in Chapter 3). Given this interaction it is likely that the underlying representations are multimodal at some level. Therefore, throughout this thesis the term 'phonological representations' will be used to refer to multimodal sublexical representations. With reduced access to auditory information it is likely that deaf children may use the other sources in order to establish representations of the sublexical structure of spoken words. These representations are unlikely to ever be as rich or as fine-grained as those established by hearing children. Nevertheless, these other sources of information may be sufficient to establish some level of phonological representation, albeit to varying degrees for individual children. It is likely that hearing children also make use of these other sources of information in order to establish robust multimodal phonological representations. However, the weighting of each of the sources of information may differ between deaf and hearing children. This section will discuss these other possible sources of information that children may use about the sublexical structure of words.

As discussed above, orthographic information may provide deaf individuals with important additional information about the patterns in spoken language beyond what they can gain from residual hearing (Campbell & Wright, 1988). However, in non-transparent alphabetic languages, such as English, the orthographic information does not show a direct correspondence with the spoken structure of the words. This means that speech and orthographic information can provide incongruent information about the sublexical structure of words.

Visual speech information may also contribute to phonological representations and be of particular importance for deaf readers. Jerger, Damian, Spence, Tye-Murray and Abdi (2009) investigated the interference from visual speech information on a picture-naming task. They asked hearing children and children with mild-to-profound hearing loss (5-12 years old) to name pictures on a t-shirt and presented auditory-only or audiovisual (t-shirt model's face was seen talking) distractors. The distractors either started with the same sound or conflicted in place of articulation or voicing. For example, the word 'pizza' could be paired with distractors 'peach', 'teacher' or 'beast' respectively. They found that the hearing children showed an interference effect, meaning they were slower to name the pictures in the conflicting conditions ('teacher' or 'beast') than when the words matched in the initial phoneme ('peach') for both auditory-only and audiovisual presentation. The deaf children showed the interference effect for the audiovisual condition only. Jerger et al. (2009) suggested that deaf children's phonological representations are structured based on visual speech information. Speechreading, the perception of visual-only speech information, is highly associated with reading ability and with phonological awareness in deaf children, which will be discussed in Chapter 4. Elliott, Braun, Kuhlmann and Jacobs (2011) highlight that studies addressing the role of phonology in reading in deaf individuals are often unspecific about the term phonology. They argued that deaf readers may have viseme representations rather than phoneme representations. Visemes are a set of speech sounds that look the same (e.g. /m/, /p/, and /b/). Although there are fewer visemes than phonemes they still provide important information about the structure of spoken language. Elliott et al. (2011) gave deaf adults a lexical decision task with prime words presented that were pseudo-homovisemes. The pseudo-homovisemes were generally pseudohomophones (e.g. 'brane' for 'brain') but care was taken to remove control words that might visually match a real word even though it did not match in sound (e.g. 'bicks' for 'mix'). They found that participants were slower to respond to the pseudo-homovisemes but not for spelling control or unrelated control words. This supports the idea that deaf adults are able to access phonological information when reading and that visual speech information may contribute to these phonological representations.

Another possible source of phonological information is articulation. Performance on phonological awareness tasks has been associated with speech intelligibility levels (Campbell & Wright, 1988; Hanson & Fowler, 1987; Johnson & Goswami, 2010). For example, Johnson & Goswami (2010) found that speech intelligibility in a group of deaf children (5-15 years old) was associated with performance on phoneme- and rhyme-judgement tasks and suggested that articulatory skills may help to develop phonological representations.

There are several sources of information that may feed into phonological representations for deaf children, including auditory and visual speech information, articulation and orthography. Although hearing children have full access to auditory speech information to build their phonological representations, speech is by nature multimodal. Therefore, it is possible that hearing children too have multimodal phonological representations drawing from the same sources of information as deaf children. As discussed above, in hearing children, as well as in deaf children, learning to read improves phonological awareness skills, supporting the idea that orthographic information contributes to phonological representations. Similarly, an individual's own articulation may also contribute to their phonological representations. For example, Carroll, Snowling, Hulme and Stevenson (2003) measured 4-year-old hearing children's articulation skills by identifying the number of consonants correct in a picture-naming task and their phonological awareness using a variety of tasks including phoneme deletion and matching words on their initial sound. They found that children's performance on the articulation task predicted their phonological awareness skills 8 months later, suggesting that early speech skills may contribute to later phonological awareness. The contribution of visual speech information to phonological representations in hearing children will be discussed in Chapter 7. For hearing children, it is likely that auditory information is more heavily weighted than the other sources of information, but that having multiple sources of phonological information allows children to build robust representations.

2.4 Chapter summary

The role of phonological awareness in reading development in hearing children has been demonstrated in concurrent, longitudinal and training studies. Deaf children tend to show poorer performance on phonological awareness tasks than their hearing peers. In addition, it is widely debated whether or not phonological awareness is important for reading in deaf children as they have reduced access to spoken language to form robust phonological representations. However, it is likely that for both deaf and hearing children phonological representations are multimodal, including auditory and visual speech information, articulation and orthography. The next chapter will discuss visual speech perception as a source of phonetic information and how this develops in deaf and hearing children.

3 Visual speech perception

One primary aim of this thesis is to examine the role of speechreading in the development of phonological representations and reading in both hearing and deaf children. All hearing children experience audiovisual speech, and many deaf children today also have access to at least some level of auditory speech, so that speech exposure for them also is to some extent audiovisual. This chapter will describe the development of visual speech perception in both hearing and deaf children. In this chapter, the term visual speech perception refers to the use and influence of visual information in audiovisual speech perception. This includes interference paradigms, such as the McGurk effect (see below), and speech perception in noise. In contrast, the term speechreading is used to refer to visual speech perception in the absence of sound.

3.1 Hearing children

Although speech is generally considered within the auditory domain only, visual information is important for guiding speech perception. Visual speech information helps to form robust speech representations, allowing a perceiver to disambiguate noisy auditory signals. It is well established that when visual speech is congruent with auditory speech it can enhance speech perception in adults (Lusk & Mitchel, 2016; Mitchel & Weiss, 2014; Sumby & Pollack, 1954), while incongruent visual information can disrupt it (McGurk & MacDonald, 1976). For example, in the McGurk effect a visually presented /ga/ that is synchronous with an aurally presented /ba/ is perceived as a /da/. However, it is important to understand whether the same is true in children, whether this changes with development, and whether it affects language development.

Several studies have suggested that children and infants as young as 2 months old can combine auditory and visual speech information (Kuhl & Meltzoff, 1982; Patterson & Werker, 1999, 2003). Although visual information is clearly used by young infants, others have suggested that visual influence on speech perception changes throughout development (Desjardins, Rogers, & Werker, 1997). Specifically, it seems that visual speech information is more influential in adults' speech perception than children's (Massaro, 1987). Children from 3 to 8 years old perceive the McGurk effect, although less reliably than adults (McGurk & MacDonald, 1976), as do infants as young as 4 1/2 months old (Burnham & Dodd, 2004). In line with this, Havy, Foroud, Fais and Werker (2017) found that 18-month-olds were able to learn words in visual and auditory modalities but did not show cross-modal transfer of the labels of items, whereas adults did. Sekiyama and Burnham (2008) found that susceptibility to the McGurk effect was low in 6-year-olds, but increased between 6 and 8 years of age in English-speaking children, suggesting that the influence of visual information in speech perception increases with age. Jerger, Damian, Spence, Tye-Murray and Abdi (2009) showed that 5-to-9-year-old children were less distracted by visual information during a cross-modal picture-word matching task than both 4-year-olds and 10-to-14-year-olds. This supports the idea that visual influence on speech perception increases in childhood but suggests that there may be a U-shaped developmental trajectory. In each of these studies, the children were grouped into age bands rather than task performance being correlated with age. Therefore, it is difficult to specify the developmental trajectory of visual influence in speech perception, but overall the influence appears to increase with age.

It is clear that infants can attend to visual speech information from an early age and that they can combine it with auditory speech information. Visual and auditory speech information, by nature, provide highly overlapping information, which may suggest that attending to the visual information is not necessary. However, the redundancy between the two modalities is instead likely to be useful, and, as described above, visual information can enhance speech perception in adults, particularly in noisy environments. Gogate, Walker-Andrews and Bahrick (2001) suggested that being able to detect the redundancy between audiovisual speech cues may help infants to distinguish between similar sounding spoken words. Weikum et al. (2007) found that infants (age 4-6 months) can discriminate two languages based on visual-only speech. This suggests that infants can attend to the visual cues from a speaker and can use this information to discriminate between languages, which may mean they can use the visual information to track auditory speech. Erdener and Burnham (2013) found that beginner readers (5-8 years old) who were more influenced by visual speech information in a McGurk-like task were better at discriminating native speech sounds than those who were less influenced by the visual speech information. They tentatively suggested that enhanced attention to visual information at this time may aid children to form grapheme-phoneme correspondences for reading acquisition but called for more direct evidence for this theory.

As visual speech information can help infants distinguish between sounds and words it is likely that it has an influence language development. Visual speech perception influences spoken word recognition in infants, suggesting that it plays a role in language acquisition (Weatherhead & White, 2017). Jerger, Damian, McAlpine and Abdi (2018) found an association between visual speech perception skills and receptive vocabulary ability for children between 4 and 14 years old. However, Erdener and Burnham (2018) found that auditory speech perception at 3 years old but not visual-only speech perception, predicted receptive vocabulary at age 4. They suggested that visual-speech perception is related to language development only in infants and at the onset of reading, but perhaps not for 3-4-year-olds. The role of visual speech perception in language development for hearing children will be discussed further in Chapter 7.

Overall, there is some evidence that visual speech perception plays a role in spoken language acquisition, supporting hearing children's perceptual discrimination of language sounds and potentially supporting the development of receptive vocabulary. The influence of visual information in auditory-visual speech perception changes throughout development.

3.1.1 Speechreading

In hearing children, the ability to make use of visual speech information from audiovisual speech signals is highly related to speechreading skill; children who show greater benefits from visual cues from speech are also better at identifying speech from purely visual speech signals (e.g. Erdener & Burnham, 2018; Erdener & Burnham, 2013; Jerger, Damian, Tye-Murray, & Abdi, 2014; Knowland, Evans, Snell, & Rosen, 2016). Therefore, this section considers findings relating to visual speech perception without corresponding auditory information. Speechreading, with no sound, is difficult as many speech sounds, such as /g/ and /k/, are not visible on the lips. In addition, for speech sounds that are visible on the lips, there is substantial overlap in the lip patterns for consonants, for example between /m/, /p/, and /b/, making it difficult to fully access the speech signal through vision alone.

Speechreading ability varies considerably among hearing adults (Jiang, Auer, Alwan, Keating, & Bernstein, 2007). There is debate as to whether speechreading ability can be trained. Some suggest it is a hard-wired skill (Montgomery & Sylvester, 1984; Summerfield, 1992) but others have shown moderate improvements in speechreading following training with adults (Bernstein, Auer, & Tucker, 2001; Blumsack, Bower, & Ross, 2007; Dodd, Plant, & Gregory, 1989; Gagne & Dinon, 1991; Gesi, Massaro, & Cohen, 1992; Massaro, Cohen, Gesi, Heredia, & Tsuzaki, 1993; Walden, Erdman, Montgomery, Schwartz, & Prosek, 1981; Walden, Prosek, Montgomery, Scherr, & Jones, 1977). The variability seen in speechreading skill is related to a number of different factors, including the individual's vocabulary skills (Lyxell & Holmberg, 2000), verbal short-term memory (Lyxell & Ronnberg, 1993), reading level (Mohammed, Campbell, MacSweeney, Barry, & Coleman, 2006), age (Tye-Murray, Sommers, & Spehar, 2007) and experience with speechreading (Bernstein, Demorest, & Tucker, 2000). Good speechreading also relies on optimal conditions, such as being able to see the full face of the speaker, no obstruction of the mouth from a hand or beard, good lighting on the speaker's face and contextual information.

Hearing children as young as 19-36 months are able to match silently spoken words to pictures (Dodd, 1987) and similar results have been found with 2-5-

year-olds (Davies, Kidd, & Lander, 2009). In Davies et al. (2009) they found no effect of age on speechreading, although this is perhaps not surprising given the limited age range in this study. Several other studies have shown that speechreading ability increases with age for hearing children, but plateaus as children get older (Dodd, McIntosh, & Woodhouse, 1998; Kyle & Harris, 2010). The nature of the developmental trajectory of speechreading skill in hearing children is not clear. While some suggest that a plateau in development occurs between 5 and 6 years of age, Dodd et al. (1998) and others suggest that it occurs later, at around 10 years of age (Kyle & Harris, 2010). In the development of the Test of Child Speechreading (TOCS) Kyle, Campbell, Mohammed, & Coleman (2013) noted that the observed plateau might be due to a lack of sensitivity of speechreading measures in the older age groups. They found a steady increase in speechreading ability from 5 to 14 years in hearing children. They also found that hearing children found it easier to speechread single words than sentences or stories, supporting previous findings (Green, Green, & Holmes, 1981). Although as a group children's speechreading improves with age, there is still huge variability in speechreading ability at every age group including adults (Kyle et al., 2013).

3.2 Deaf children

It is clear that visual information plays an important role in audiovisual speech perception for hearing adults and children, but it is of particular importance for individuals who are deaf. Having introduced audiovisual speech perception and speechreading skills in hearing children the following section will now consider speechreading in deaf adults and children only.

Even with a cochlear implant deaf children have reduced access to auditory speech signals compared to hearing children. Thus they are more dependent than hearing children on visual speech perception to aid spoken language perception. In addition, speechreading skill has been shown to enhance the effectiveness of hearing aids (Arnold, 1997) and cochlear implants (Bergeson, Pisoni, & Davis, 2005). However, as described above, many speech sounds are visually indistinguishable. Whether an individual's speechreading ability is fixed or can be improved is controversial. As described above, studies with hearing children suggest that their speechreading ability improves with age and that they find it easier to speechread single words than sentences or stories (Dodd et al., 1998; Green et al., 1981; Kyle et al., 2013; Kyle & Harris, 2010). Each of these studies also included deaf children of the same age, and found similar results with both deaf and hearing children. Deaf adults who have been deaf from an early age have equivalent or superior speechreading skills compared to hearing adults (Bernstein et al., 2000; Mohammed et al., 2006). Some studies have suggested that deaf children and adolescents have superior speechreading skills to their hearing peers (Kyle & Harris, 2006; Lyxell & Holmberg, 2000). Kyle & Harris (2006) compared deaf (7-8 years old) and hearing children (5-8 years old) matched on reading level and found that the deaf children were better at silent video-to-picture matching than the younger hearing children. Given that speechreading skill seems to develop with age in deaf children the difference in speechreading skill in this study may be due to age differences. However, Lyxell and Holmberg (2000) found that moderately deaf adolescents were better than age-matched hearing adolescents (12 years old) at speechreading single words and sentences. In the standardisation of the Test of Child Speechreading (TOCS; Kyle et al., 2013) the deaf and hearing children (5-15 years old) were matched on age and the deaf children did not have superior speechreading skills compared to their hearing peers. In addition, a study in deaf adults with cochlear implants indicated that later implantation may be associated with better speechreading scores (Pimperton, Ralph-Lewis, & MacSweeney, 2017). These results suggest that more experience with and attention to speechreading can enhance speechreading proficiency.

3.3 Chapter summary

Visual speech, in addition to auditory speech, is a potential source of information that may contribute to the development of phonological representations. Visual and auditory speech are synchronous and highly overlapping sources of information. In noisy environments visual speech information can enhance speech perception and it has been shown that incongruent visual and auditory information can disrupt speech perception as in the McGurk effect. Visual speech information can help hearing infants distinguish between sounds and words and is therefore likely to play a role in language acquisition.

Speechreading ability relates to the influence that visual information has in audiovisual speech perception. However, speechreading is a difficult skill because many speech sounds overlap in their lip patterns (e.g. /m/, /p/ and /b/). Levels of speechreading skill are highly variable in both deaf and hearing groups. Speechreading is of particular importance to deaf individuals, who have reduced access to auditory speech information. Although there is debate about whether speechreading can be trained, deaf adults, who have had a lifelong reliance and experience with speechreading, tend to be better than hearing adults on speechreading tasks, suggesting that it improves as a result of extensive practice. In addition both deaf and hearing children's ability to speechread seems to develop with age, supporting the idea that it has the potential to change. The next chapter will discuss how speechreading may relate to phonological awareness and reading ability in both deaf and hearing children.

4 The relationship between speechreading, phonological awareness and reading in deaf and hearing children

In the previous chapters the development of reading, phonological awareness and speechreading have been summarised. This chapter will outline how these three factors may relate to each other in both deaf and hearing children. It will then summarise Kyle's (2015) model of reading in deaf children, which brings these factors together to explain how speechreading may influence the development of reading in deaf children. Finally, the Speechreading Training and Reading intervention (Pimperton et al., submitted) that formed the basis for this thesis will be summarised.

Audiovisual speech perception skills have been shown to relate to reading ability in hearing adults, with poorer readers showing deficits in audiovisual speech integration (Francisco, Groen, Jesse, & McQueen, 2017; Francisco, Jesse, Groen, & McQueen, 2014). In addition, hearing dyslexic adults tend to show poorer speechreading skills than hearing adults without a history of dyslexia (de Gelder & Vroomen, 1998; Mohammed et al., 2006). However, there does not seem to be a relationship between speechreading and reading ability in hearing adults without a history of dyslexia, though this may be due to a lack of variation in reading scores reducing the ability to detect a correlation (Mohammed et al., 2006). Importantly though, for both deaf and hearing dyslexic adults there is a relationship between speechreading and reading (r = .58 and r = .54 respectively, Mohammed et al., 2006).

Speechreading skill correlates with reading in deaf children (Arnold & Köpsel, 1996; r = .49, Kyle, Campbell, & MacSweeney, 2016; r = .60, Kyle & Harris, 2006) regardless of the child's preferred language (Kyle & Harris, 2010, 2011); across linguistic levels (single word and sentences) (Kyle et al., 2016); and is a longitudinal predictor of reading development (r = .53, Harris, Terlektsi, & Kyle, 2017; r = .64, Kyle & Harris, 2010). Perhaps surprisingly, speechreading also relates to reading accuracy in hearing children (r = .31, Kyle et al., 2016; r = .58, Kyle & Harris, 2010). Speechreading is an independent predictor of reading ability when controlling for vocabulary skills in both deaf and hearing children, accounting for 11% of the variance in deaf children's reading scores (Kyle et al., 2016).

Several researchers have suggested that speechreading and reading in deaf and hearing children may be related as speechreading provides one of the sources of information for developing phonological representations (Leybaert, 1993; Mohammed et al., 2006; Woll, 2012). Written language for alphabetic languages represents the spoken word patterns, which contain both auditory and visual information. As described in Chapter 2, phonological representations of spoken language are likely to be multimodal, representing what a person hears and sees when someone speaks as well as their own articulation of speech sounds. For deaf children, the importance of the different sources of speech information is likely to be weighted away from the degraded auditory signal with a greater weight being given to visual speech information. Indeed, several authors have suggested that deaf children develop their phonological awareness via speechread information (Campbell & Dodd, 1980; Kyle, 2015; Kyle & Harris, 2011; Woll, 2012). Dodd and Hermelin (1977) found that adolescents born deaf primarily used speechread information to complete a series of phonological matching tasks (e.g. rhyme judgement), as opposed to orthographic, lexical or motor information. Dodd, Hobson, Brasher and Campbell (1983) found that deaf adolescents (13-16 years old) showed recency effects in speechread serial recall tasks but not orthographic serial recall tasks, suggesting that speechread information is phonologically coded in a similar way to auditory speech information. Deaf children's spelling errors also give insight into their phonological coding, with many spelling errors being an approximation of speechreading for orally educated deaf children (Burden & Campbell, 1994; Hanson, Shankweiler, & Fischer, 1983; Leybaert & Alegria, 1995; Sutcliffe, Dowker, & Campbell, 1999). This supports the idea that speechreading is an important source of information for the development of phonological representations in deaf children.

Recent studies have suggested that speechreading is correlated with performance on more explicit phonological awareness tasks, such as rhyme judgements (Campbell & Wright, 1988), both concurrently ($r = .46$; Kyle & Harris, 2006) and longitudinally (Kyle & Harris, 2010) in deaf children. In deaf adults, similar relationships have been found between speechreading, phonological awareness and reading (Mohammed et al., 2006). However, when phonological awareness was controlled for, the relationship between speechreading and reading remained ($r = .49$). This suggests, that although speechreading relates to phonological awareness skills, there may be other factors contributing to the relationship between speechreading and reading in deaf adults.

The idea that visual information can be used to develop phonological representations is supported by evidence from children using Cued Speech (Leybaert & Alegria, 2003; Leybaert, Bayard, Colin, & LaSasso, 2015). As described in Chapter 1, Cued Speech is a system of manual gestures used to accompany spoken language to disambiguate speechread phonetic information. Children who use Cued Speech tend to develop well-specified phonological representations, with better phonological awareness and reading skills than their deaf peers who do not use Cued Speech (Bouton, Bertoncini, Serniclaes, & Cole, 2011; Charlier & Leybaert, 2000; Crain & LaSasso, 2010) as they have full access to the phonetic information in spoken language. Deaf children who use Cued Speech also tend to have comparable reading ability to their hearing peers (Colin, Leybaert, Ecalle, & Magnan, 2013; Crain & LaSasso, 2010; Leybaert, 2000; Rees & Bladel, 2013) when exposed to Cued Speech before 3 years of age. This suggests that rather than phonological representations being solely auditory, visual information can also be used to develop them and that it is the quality of information rather than its modality that is important. In addition, these findings are in line with the idea that phonological representations are important for developing good reading skills.

Visual speech information is clearly important for deaf children to develop representations of spoken words and, although less considered, is also important for hearing children. Teinonen, Aslin, Alku and Csibra (2008) found that 6-month-old hearing infants could use visual speech information to enhance the distinction between phonetic contrasts, such as /ba/ and /da/. This support from visual information in phonetic discrimination also aided the infants' learning of new categories, which they were able to transfer onto auditory-only test items. Visual and auditory speech information are complementary, as phonemes that are hard to distinguish by sound are easy to distinguish visually (e.g. /n/ and /m/) and vice versa (e.g. /m/, /p/ and /b/) (Campbell, 2011; Woll, 2012).

However, conflicting results have been found regarding the relationship between speechreading, phonological awareness and reading in hearing adults with dyslexia. Mohammed et al. (2006) found that, as with deaf adults, hearing adults with compensated dyslexia showed a positive relationship between speechreading and reading skill. They suggested that poorer speechreading in the hearing dyslexic group may be explained by a deficit in phonological representations, which was supported by the finding that when phonological awareness scores (measured by a sound-matching task, e.g. saying whether a pair of words rhymed or not) were partialled out, the relationship between speechreading and reading disappeared. Conversely, Francisco, Groen, Jesse and McQueen (2017) found that in a group of dyslexic adults, better speechreading (identifying consonants from silent videos) was associated with lower phonological awareness scores (measured with a sound-reversal task, e.g. whether a spoken word 'tac' was the reversal of a second word 'cat'). They suggested that the dyslexic adults with lower phonological skills relied more on visual information as a compensatory strategy. It is not clear why these studies show such discrepant results, although the measures of speechreading and phonological awareness differed considerably between the two studies.

4.1 Model of reading for deaf children (Kyle, 2015)

The ideas discussed above were summarised by Kyle (2015) in the model of reading in deaf children (Figure 2) based on the simple view of reading (Gough & Tunmer, 1986). In Kyle's model, vocabulary and speechreading relate directly to reading ability, and phonological awareness is built up through both reading and speechreading. As phonological awareness becomes established, it begins to predict reading ability and the two skills have a reciprocal relationship, as shown below.

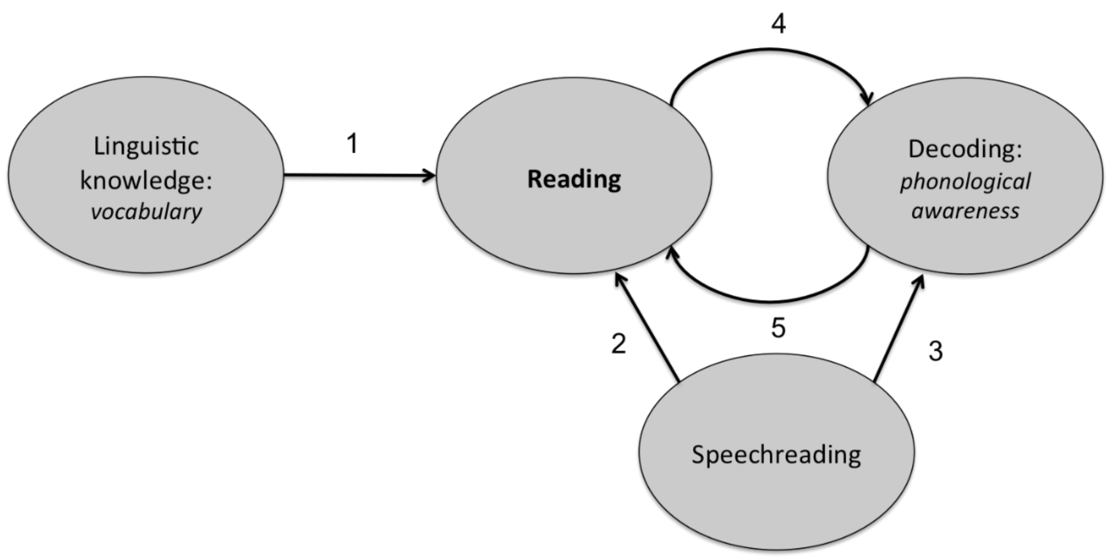

Figure 2. Kyle's (2015) model of reading in deaf children.

It is likely that the nature of the relationship between each element of this model is bidirectional. As well as speechreading contributing to phonological awareness, as discussed above, an individual with better phonological awareness is likely to be able to use their phonological representations to support access to the speechreading signal. This idea is in line with the finding that dyslexic adults have poorer speechreading skills than non-dyslexic adults as poorer speechreading may reflect the poorer phonological skills that characterise the dyslexic group. In addition, in Mohammed et al.'s (2006) study the relationship between speechreading and reading in dyslexic adults was not significant when phonological awareness was controlled for. This may indicate that phonological awareness was the limiting factor in performance on both the reading measures and the speechreading measures.

Similarly, the relationship between phonological awareness and reading may be reciprocal in both hearing (e.g. Nation & Hulme, 2011) and deaf (e.g. Kyle & Harris, 2010) children. As a child learns to read, they can use orthography to further support phonological development. This may be especially true for deaf children, who have reduced access to phonological information through speech and it has been shown that reading at age 8 predicts phonological awareness in deaf children at age 10 but not the other way around (Kyle & Harris, 2010).

Vocabulary is a very important predictor of reading in both deaf and hearing children, reflecting the comprehension aspect of the simple view of reading. However, this thesis will focus on decoding to investigate the relationship between speechreading, phonological awareness and reading. As discussed above, speechreading also relates to reading ability in hearing children, and therefore the applicability of this model to hearing children’s reading will also be investigated.

4.2 Summary

Reading is an incredibly important skill for children to learn in the early stages of school in order for them to access the rest of education. Research with hearing children suggests that both decoding and language skills are important for the development of reading comprehension. Decoding depends on several skills including phonological awareness, which is thought to be impaired in developmental dyslexia. Although phonological awareness is important for reading development in hearing children there is some debate as to whether the same is true for reading development in deaf children. In order to develop robust representations of the phonological structures in words it is important to be able to have full access to that phonological information. Deaf children have reduced access to auditory speech information and so are likely to have poorer phonological representations than their hearing peers, which is one factor that is likely to contribute to poorer reading skills. However, although phonology is often used in reference to the sound structures within words, speech is by nature multimodal and there are many additional sources of phonological information including what we see when someone is talking and our own articulation. Visual speech information may contribute to phonological representations and speechreading ability has been shown to relate to both phonological awareness and reading in deaf children. Kyle's (2015) model of reading in deaf children summarises the potential relationship between speechreading, phonological awareness and reading. Interestingly, visual speech perception also relates to reading ability and language development in hearing children, suggesting that this model might also apply to reading in hearing children.

4.3 The Speechreading Training and Reading Intervention

In response to the growing evidence that speechreading relates to phonological awareness skills and reading ability in young deaf children, Pimperton et al. (submitted) investigated whether speechreading can be trained in young deaf children. They conducted a single-blind randomised controlled trial using a computerised speechreading training (STAR) programme to improve speechreading skills. Secondary questions were whether improvements in speechreading would lead to secondary benefits in phonological awareness and reading. The Speechreading Training and Reading intervention (referred to from here as STAR_D) formed the foundation of my PhD work. New data collected in this thesis from hearing children using the same intervention will be referred to as STAR_H. The STAR_D project was funded by a Wellcome Trust Fellowship. I joined the project for the final stage of testing (Time 4 follow-up).

The resulting paper from the STAR_D project (Pimperton et al., submitted) is presented in Appendix A. The children in this project were assessed on a range of cognitive and language tests before the start of the training (baseline) and then at three follow-up timepoints: immediately after the training, 3 months post-intervention and 10 months post-intervention. The pre- and post-tests included assessments of speechreading, reading, phonological awareness, vocabulary, numerical skills and non-verbal IQ, which are described in detail in the paper.

The intervention involved playing a speechreading and reading computer game set in space for 10 minutes a day, 4 days a week for 12 weeks. The control group played the same games with number and maths content. The games were adaptive based on the child's performance.

The primary outcome measure was the Test of Child Speechreading (TOCS; Kyle, Campbell, Mohammed, & Coleman, 2013). This measures speechreading of single words, sentences and stories with multiple-choice picture options at each level. There was no significant difference between the speechreading intervention group and the control group as a result of the intervention on this task. However, there was a large effect of training on the number of questions correctly identified in the TOCS extension Everyday Questions subtest, three months post-intervention. The TOCS extension involves speechreading single sentences and repeating back each sentence. This effect was moderate at the immediate post-test and 10 months after the intervention.

In addition, there was a moderate effect of training on both phonological representations and phonological awareness scores at the three-month post-test, although there was no effect on reading scores.

These results provide some evidence that speechreading can be trained in young deaf children, and that, for some deaf children, this can have a knock-on effect on their phonological representations and awareness. This thesis therefore aimed to further understand the relationship between speechreading and reading in deaf children and the role of phonological awareness in the relationship. The goal was to address the question 'what is the role of speechreading in reading for deaf children?'. In addition, it aimed to investigate whether the observed relationship between speechreading,

reading and phonological awareness in deaf children was reflected in hearing children despite the fact that they have lower reliance on visual speech perception than their deaf peers.

4.4 Thesis overview

This thesis will address the following questions:

- **How do eye movements during speechreading relate to speechreading skill in deaf and hearing children?**

Chapter 5 uses eyetracking during a sentence speechreading task in deaf and hearing children to investigate whether they show similar eye-movement patterns to each other whilst watching videos of silent speech. We also addressed whether the time spent looking at the mouth during these silent videos correlates with their speechreading ability. Similar eye-movement patterns, we argue, would suggest that deaf and hearing children access visual speech in a similar way and therefore they may be able to use visual speech information in the same way to contribute to spoken word representations.

- **Is the relationship between speechreading and reading in deaf children mediated by phonological awareness?**

This question is investigated in Chapter 6 using structural equation modelling on the data from the pre-training assessments of deaf children in the STAR_D study.

- **Is the relationship between speechreading and reading in hearing children mediated by phonological awareness?**

To address this question, in Chapter 7 new data from young hearing children (5-8 years old) were collected and analysed using structural equation modelling.

- **Can speechreading be trained in hearing children and does this lead to improvements in phonological awareness and reading?**

To test the model derived in Chapter 7, a trial of the STAR training programme that had been used with deaf children was carried out with young hearing children (4-5 years old).

5 Visual speech perception in deaf and hearing children

The study reported in this chapter has already been published. The report of the study below has been adapted from the paper to fit with this thesis.

Reference: Worster, E., Pimperton, H., Ralph-Lewis, A., Monroy, L., Hulme, C., & MacSweeney, M. (2018). Eye Movements During Visual Speech Perception in Deaf and Hearing Children. *Language Learning*, (June), 159–179. http://doi.org/10.1111/lang.12264

As introduced in Chapter 4, speechreading is related to reading ability in both deaf and hearing children. Chapter 1 noted that in general deaf children have poorer reading ability than their hearing peers and Chapter 4 introduced the idea that visual speech perception abilities may link to language development in hearing as well as deaf children. Given the relationship between speechreading and reading in deaf and hearing children, it is important to understand what makes a good speechreader. Although speechreading skill is highly variable between individuals, it is not clear what accounts for this variation or what strategies may be advantageous. Gaze direction during visual speech perception has not been studied widely in deaf adults or children and may provide insights into underlying processes used by children during speechreading.

Some studies have shown that hearing adults from western cultures tend to look at the eyes more than the mouth during audiovisual speech perception in quiet (Smith, Gibilisco, Meisinger, & Hankey, 2013; Vatikiotis-Bateson, Eigsti, Yano, & Munhall, 1998), while others have observed a preference for the mouth over the eyes (Barenholtz, Mavica, & Lewkowicz, 2016). These differences between findings may reflect the fact that the allocation of visual attention to the face during speech perception varies as a function of task demands. Lansing and McConkie (1999) showed that participants allocate their attention towards the eyes when identifying emotional or prosodic information from audiovisual speech and towards the mouth when the task emphasises segmental information. This suggests that viewers may allocate their attention to the most useful source of information. As well as being task-dependent, gaze shifts as noise in the auditory stream increases, with participants focusing further down the face to the nose and mouth (Vatikiotis-Bateson et al., 1998). This suggests that as speech perception becomes more difficult, perceivers are able to shift their attention to the mouth to make use of visual cues. Barenholtz et al. (2016) found that looking time to the mouth is modulated by language familiarity, with adults looking longer at the mouth when watching an unfamiliar language than a familiar one. The mouth is clearly an important source of information in audiovisual speech perception, especially when segmentation of speech is a priority or when there is uncertainty about the speech signal.

To date, however, there has been relatively little research investigating whether there is a relationship between gaze patterns and performance in audiovisual speech perception. That is, whether looking to the mouth (mouth-focus) provides an advantage in audiovisual speech perception. In conditions of no noise with a single speaker, fixating away from the mouth, up to 15 degrees of eccentricity, does not affect audiovisual speech intelligibility (Yi, Wong, & Eizenman, 2013). Similarly, the McGurk effect is still observed when participants fixate 10 degrees from the centre of the mouth (Paré, Richler, ten Hove, & Munhall, 2003). These results suggest that visual speech information can be accessed via peripheral vision, as suggested by Massaro (1998), calling into question why individuals fixate the mouth in difficult perceptual situations. However, when noise is introduced by having two speakers presented side-by-side, speech intelligibility scores are reduced when fixating more than 2.5 degrees of eccentricity from the centre of the mouth (Yi et al., 2013). This suggests that as auditory speech perception becomes more difficult, peripheral vision is not sufficient to access supporting visual speech information from the mouth. It seems from these results that mouth-focus does aid audiovisual speech perception. However, studies with hearing adults have found that mouth-focus does not correlate with individuals' ability to speechread silent spoken sentences (Lansing & McConkie, 2003), to speechread consonant-vowel-consonant clusters (Wilson, Alsius, Paré, & Munhall, 2016) or relate to susceptibility to the McGurk effect (Paré et al., 2003). These results are surprising given that the gaze shifts to the mouth with increasing perceptual difficulty during audiovisual speech perception. However, each of these studies only had 20 participants or fewer. Thus the lack of correlation may be due to a lack of statistical power.

Although no developmental research has directly related gaze to the mouth to performance on speech perception tasks, Young, Gregory, Merin, Rogers and Ozonoff (2009) found that increased gaze to the mouth at 6 months of age predicted higher expressive language outcomes 18 months later. Similarly, a case study with a deaf, skilled speechreader showed that she looked at the mouth during visual speech perception tasks (Lansing & McConkie, 1994), supporting the idea that mouth-focus improves access to phonetic information in visual speech.

As described in Chapter 3, visual influence on speech perception develops over time, increasing particularly between 6 and 8 years old (Sekiyama & Burnham, 2008). Along with changes in visual influence on speech perception, there are also developmental changes in gaze patterns to the face. Infants younger than 8 months old show a preference for watching the eyes both during infant-directed and adult speech (Lewkowicz & Hansen-Tift, 2012; Smith et al., 2013). At around eight months old, infants shift their attention to the mouth of a speaking face but return to focus on the eyes again by 12 months old (Lewkowicz & Hansen-Tift, 2012; Pons, Bosch, & Lewkowicz, 2015). In addition bilingual infants shift to watch the mouth earlier than monolingual infants, showing equal looking to the eyes and mouth at 4 months old, and maintain their preference for the mouth at 12 months old (Pons et al., 2015). For bilingual infants it is likely that visual information from the mouth aids differentiation between the two languages they are acquiring and thus supports language acquisition.

Overall, the developmental evidence is consistent with findings from adults that as speech perception becomes harder or more important, gaze to the mouth is prioritised. This suggests that the mouth is an important source of speech information and that children can selectively allocate attention to make use of this information. In the current study we investigated whether children who are born deaf access visual speech in a different way from hearing children. This is a possibility since deaf children are likely to have experienced a greater dependence on visual speech throughout their lifetime. We used eye tracking with children born moderately-to-profoundly deaf and hearing children aged 5-8 years old while they watched videos of silently spoken sentences. We aimed to address three questions: 1) Do deaf and hearing children differ in the time spent looking at the mouth during speechreading? 2) Does the time spent looking at the mouth during speechreading relate to speechreading ability in deaf and hearing children? 3) Does the above relationship differ between deaf and hearing children?

5.1 Methods

5.1.1 Participants

The deaf children in this study were a subset of 33 children (20 males) from the STAR_D project (17 experimental group, 16 control group; see Appendix A). In this subset the children had an average bilateral hearing loss of 93.71db in the better ear (37.5db – 120db). Twelve children had cochlear implants bilaterally, 18 had hearing aids bilaterally, two children had no aiding and one child had one hearing aid and one cochlear implant. Six of the children used only British Sign Language in the classroom, 14 of them used a mixture of speech and sign and 13 of them used spoken English only. These studies were conducted simultaneously with the 10-month follow-up of the STAR_D project. At this timepoint the children had a mean age of 7 years and 2 months (*SD* = 7.6 months). Eyetracking calibration was not possible with two deaf children who therefore could not be tested further. A further two children were excluded due to having fewer than 8 trials with more than 50% tracking. This left 29 participants (18 males). Fifteen of the children in the current study were in the speechreading training group in the STAR project and 14 were in the maths training group (control group). They had a mean age of 7 years and 2 months (*SD* = 7.7 months).

A control group of 59 hearing children (32 males) was recruited from two mainstream schools in Cambridgeshire. Twenty-nine children (18 males) were selected from this group to be matched to the deaf children in age and gender. Of these 29, 21 children were monolingual English speakers, 5 spoke an additional language at home but had learnt English from birth and the remaining 3 children had been learning English for an average of 3 years (*SD* = 2 years; range: 1-5 years), as reported by their parents. Despite this, these three children had an average T-score of 48 on the British Ability Scales Word Definitions subtest (group range: 42-51) and were therefore included in the study. The hearing group of participants had a mean age of 6 years and 11 months (*SD* = 5.8 months).

5.1.2 Offline measures

5.1.2.1 Speechreading

To assess speechreading ability, the Test of Child Speechreading was used (TOCS; Kyle et al., 2013, https://dcalportal.org/). For the TOCS words subtest the children watched 15 silent videos of a model (7 male, 8 female) speaking a single word. After each video they selected one of four presented pictures to match the word they just saw. Each child was first familiarised with both models by watching silent videos of them saying the days of the week. After familiarisation each child had three practice trials before the main test began. The task was self-paced and lasted approximately 5 minutes.

5.1.2.2 Reading

Reading was assessed using the YARC early word and single word reading subtests (Hulme et al., 2009). Children were allowed to respond in either English or BSL and were awarded a point for each item they labelled correctly.

5.1.2.3 Vocabulary

The deaf children's vocabulary knowledge was assessed using a picture-naming task. Each child was shown one picture at a time, taken from the training computer game, and asked to give the name in either speech or sign. They were given one point for each picture they could name correctly in either modality. A response was considered correct if it could be identified as the target word, regardless of pronunciation.

The hearing children were not involved in the training study. Therefore their vocabulary knowledge was assessed using a standardised measure: the British Ability Scales third edition (BAS3) Expressive Vocabulary subtest (Elliott & Smith, 2011).

5.1.2.4 Non-verbal IQ

All the deaf children had completed the BAS3 Matrices subtest at the first timepoint in the randomised control trial. This was 16 months before the current data collection point. These data are reported in Table 1. The hearing children completed the BAS3 Matrices subtest in the same testing session as when the eyetracking data were collected.

5.1.2.5 Summary of measures

The children's scores on speechreading, reading, non-verbal IQ and vocabulary are shown in Table 1. For the YARC early word reading subtest there was no difference between groups but this appears to reflect a ceiling effect in both groups. However, the hearing children had significantly higher scores than the deaf children on the more difficult YARC single word reading subtest. In addition, the hearing children had higher scores than the deaf children on the BAS3 Matrices subtest, as shown in Table 1. However, as the measurement on the BAS3 Matrices subtest was taken at different timepoints in relation to when the eyetracking was conducted and at different ages for the deaf and hearing children the scores were not considered directly comparable. Although there was a substantial difference between groups on the Test of Child Speechreading words subtest (*d* = 0.41) this difference was not significant due to low statistical power.

Table 1. Participant scores on measures of speechreading, reading, general ability and vocabulary

Test	**Subtest (max range)**	**Deaf**		**Hearing**		**t(*df*)**	***p***	***d***
		Mean (*SD*)	**Range**	**Mean (*SD*)**	**Range**			
Test of Child Speechreading	Words (0 to 15)	8.62 (*2.57*)	3-12	7.45 (*2.72*)	2-12	1.69 (*56*)	.111	0.42
York Assessment of Reading Comprehension	Early Word Reading (0 to 30)	24.00 (*7.94*)[a]	2-30	26.14 (*5.24*)	7-30	1.21 (*56*)	.238	0.31
	Single Word Reading (0 to 60)	19.10 (*10.96*)	0-53	26.72 (*13.83*)	5-55	2.34 (*56*)	.027	0.61
British Ability Scales	Matrices T-score (0 to 80)	35.90 (*6.48*)	26-48	50.86 (*12.82*)	23-79	-	-	-
	Vocabulary T-score (0 to 80)	-	-	51.69 (*7.50*)	41-68	-	-	-
Picture naming	(0 to 74)	65.93 (*3.95*)	56-74	-	-	-	-	-

n = 29.

5.1.3 Eyetracking methods

Eye movements were recorded using a RED250 eyetracker manufactured by Sensomotoric Instruments (SMI, sampling rate 250Hz). The children were seated with their head approximately 60cm from a laptop screen and tracking was accommodated between 50cm and 80cm from the screen. The children were asked to sit as still as possible. Both eyes were tracked but, as is standard practice, only data from the right eye were used. The eyetracker was first calibrated using a five-point calibration with a smiley face used as the calibration point. The child was asked to follow the nose in the centre of the face. A four-point validation was then carried out and the calibration process was repeated if necessary. Drift-correction trials were placed in between each video stimulus, showing a smiley face in the centre of the screen.

5.1.4 Online measure – *Test of Child Speechreading – Everyday questions subtest*

Eyetracking data were collected as the children performed the 'Everyday Questions' subtest of the Test of Child Speechreading (Kyle et al., 2013, https://dcalportal.org/). In this subtest each child watched 12 silent videos of a person asking everyday questions such as "How old are you?". Six were spoken by a male model and six by a female model. The child was asked to watch each video and then repeat as much of the question as they could. They could give their response in either speech or sign and were given a point for each lexica item labelled (maximum score 62).

In between each video a drift-correction screen was presented with a smiley face in the centre of the screen. The deaf children were asked to look at the smiley face before the next trial was manually triggered. For the hearing children, looking at the smiley face for 1 second automatically triggered the following trial. If it was not triggered automatically, the experimenter continued the experiment manually and made a note of the corresponding trial.

5.1.5 Data Analysis

The eyetracking data were first cleaned by ensuring that the calibration was correct on the drift-correct trials. If not, the calibration was adjusted by moving the eye marker to the smiley face, changing the calibration for all subsequent trials. Repeated trials where the child was not on-task were removed. Any trials (max = 12) where the child was looking at the screen for less than 50% of the speech time were removed. For the deaf children, the average number of retained trials was 11 (range: 9-12). For the hearing children, only one child had one trial removed and the rest were retained.

Three areas of interest (AOI) were created: one that encompassed the whole screen, one that identified the upper face (Eyes), and one that identified the lower face (Mouth). The Eyes and Mouth AOIs were created using equal-sized semicircles with the flat edge of each meeting on the nose and not overlapping. The AOIs were then moved with the video such that their meeting edge remained equidistant from the centre of the eyes and mouth on the image.

Each stimulus video was coded for the onset and offset of visual speech taken as the moments when the lips first moved from a closed position and when the lips returned to that position. The percentage Net Dwell Time (%NDT) was extracted for each of the three AOIs for each trial – for the pre-speech segment, the speech segment and the post-speech segment individually. Each video stimulus was a different length so %NDT was used to allow averaging across trials.

Regression models were used to address the hypotheses. Group (speechreading training or maths training) was dummy coded and entered into the regression models in order to test mean differences in performance between the groups.

5.1.5.1 Social Tuning Score

During analysis we noticed a consistent pattern across participants where the child started each trial by gazing at the eyes, then shifted their gaze to the mouth at the speech onset, returning to the eyes at speech offset, as shown in Figure 3.

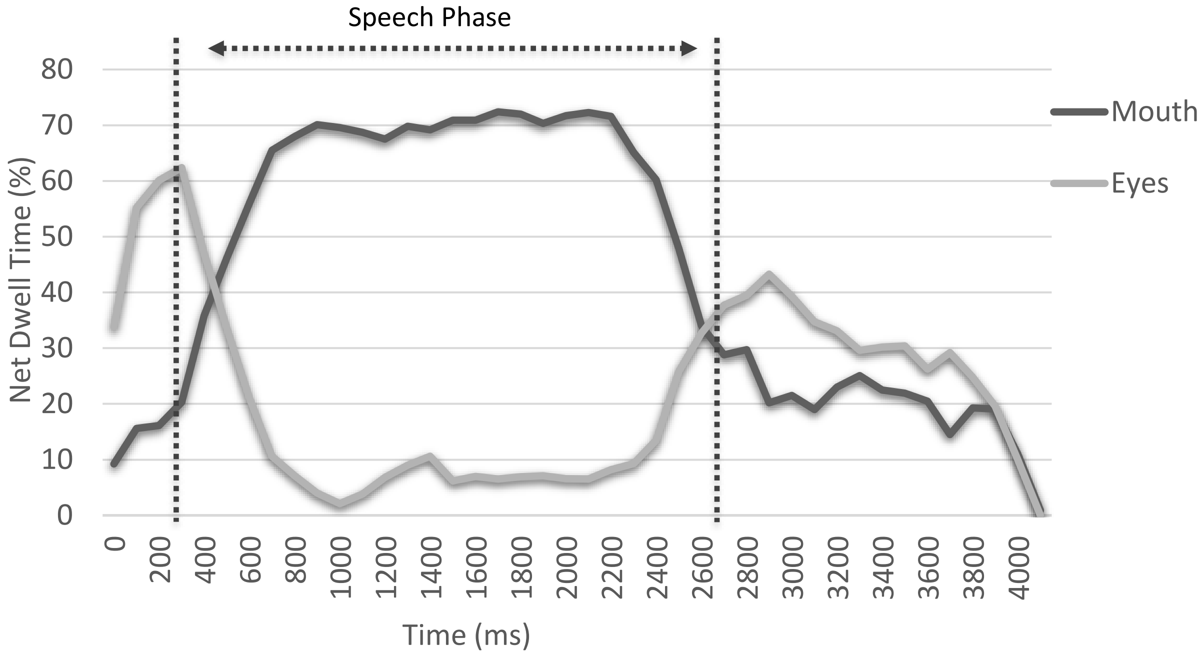

Figure 3. Percentage net dwell times averaged across participants for one trial, showing the tendency to look to the eyes before and after speech and at the mouth during speech. The dotted lines indicate the start and finish of speech.

To determine whether this strategy was advantageous for speechreading a scoring system was devised using the following formula:

$$\textbf{Ratio score} = 1/3 \left[\overset{\textbf{Pre-speech}}{\frac{\text{Time on eyes} - \text{Time on mouth}}{\text{Total Pre-speech time}}} + \overset{\textbf{Speech}}{\frac{\text{Time on mouth} - \text{Time on eyes}}{\text{Total Speech time}}} + \overset{\textbf{Post-speech}}{\frac{\text{Time on eyes} - \text{Time on mouth}}{\text{Total Post-speech time}}} \right]$$

This was used to give a measure of preference for the eyes-mouth-eyes pattern observed. This score is referred to as the social-tuning ratio from here on. A higher social-tuning ratio reflects greater use of the eyes-mouth-eyes pattern when watching the silent videos.

5.2 Results

Descriptive statistics for all offline measures are shown in Table 1 above. As the measures were not all normally distributed, bootstrapping was used in all analyses.

There was no significant difference in the number of words correctly identified in the speechreading task between the deaf (*M* = 18.72, *SD* = 14.37) and hearing participants (*M* = 17.45, *SD* = 9.39), *t*(48.2) = 0.40, *p* = .701, *d* = 0.10. There was no significant correlation between level of hearing loss for the deaf children and the number of words correctly identified in the speechreading task, *r*(18) = –.353, *p* = .127.

5.2.1 Percentage Net Dwell Time on the mouth

5.2.1.1 Group contrast of percentage Net Dwell Time on the mouth

There was no significant difference in percentage Net Dwell Time (%NDT) on the mouth during speech between the deaf (*M* = 64.29, *SD* = 16.89) and hearing participants (*M* = 70.33, *SD* = 16.22), *t*(56) = 1.39, *p* = .161, *d* = 0.36. However, for the deaf children there was a significant difference in %NDT on the mouth during speech between those who did speechreading training (*M* = 70.96, *SD* = 13.08) and those who did maths training (*M* = 57.15, *SD* = 17.99), *t*(27) = 2.38, *p* = .029, *d* = 0.88.

5.2.1.2 Relationship between speechreading scores and percentage Net Dwell Time on the mouth

There was a significant positive correlation between %NDT on the mouth during speech and the number of lexical items identified in the TOCS extension task for the deaf children ($r(27) = .399$, $p = .032$) and the hearing children ($r(27) = .586$, $p = .001$). The relationships between %NDT on the mouth and speechreading scores for both groups are depicted in Figure 4.

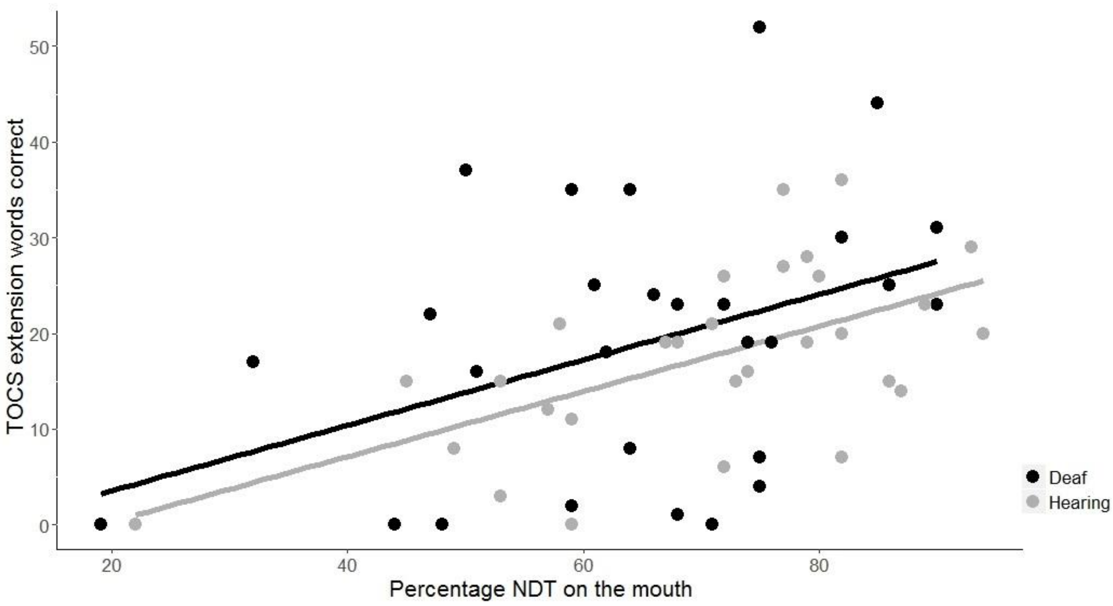

Figure 4. The number of words correctly identified in the TOCS Everyday Questions task plotted against % Net Dwell Time on the mouth for both deaf and hearing participants. The relationship between these variables was significant for both groups and it is clear that the slopes for the two groups are essentially identical.

5.2.1.3 Group differences in the relationship between speechreading score and percentage Net Dwell Time on the mouth

The relationship between speechreading score and the percentage Net Dwell Time on the mouth was significant for both the deaf and hearing groups.

From Figure 4 it is also clear that the relationship between these variables is very similar in the deaf and hearing groups.

To determine whether the relationship between mouth-focus and speechreading performance differed between the deaf participants in the speechreading and maths training groups, a multiple regression was carried out. The number of lexical items identified in the TOCS extension task was predicted from %NDT on the mouth during speech, Group and the interaction between these two variables. The participants' predicted number of lexical items identified in the TOCS extension task was equal to –5.41 + 0.35(%NDT on mouth) + 16.36(Group) – 1.78(Interaction), where Group was coded as 0 = Maths, 1 = Speechreading. None of the %NDT on mouth (p = .101), Group (p = .342) or the interaction term (p = .502) were significant predictors of the speechreading scores. When the interaction was dropped from the model the participants' predicted number of lexical items identified in the TOCS extension task was equal to –1.75 + 0.28(%NDT on mouth) + 4.71(Group). The %NDT on mouth was a significant predictor (p = .046) whilst Group (p = .434) was not. As there was no significant interaction between the speechreading and maths training groups, the deaf participants can be treated as a single group.

In summary, children in the speechreading training group looked at the mouth more (higher mouth %NDT) than those in the maths training group. However, %NDT on the mouth predicts speechreading scores to a significant and equal degree in both groups.

5.2.2 Social-tuning ratio

Having identified the social-tuning pattern during analysis we conducted exploratory analyses to test whether it relates to performance on the speechreading task.

5.2.2.1 Group differences in social-tuning ratio

We used t-tests to investigate group differences between the deaf and hearing participants and between speechreading training and maths training groups in social-tuning ratio. There was a significant group difference in social-tuning ratio with hearing children (M = .48, SD = .14) demonstrating this pattern more reliably than deaf children (M = .37, SD = .11), $t(56) = -3.34$, p = .004, d = 0.87. Within the group of deaf children, there was no significant difference in the social-tuning ratio between those who completed the speechreading training (M = .39, SD = .12) and those who completed the maths training (M = .36, SD = .10), $t(27) = -0.76$, p = .463, d = 0.27.

5.2.2.2 Predicting speechreading scores using the social-tuning ratio

There was a significant positive correlation between the social-tuning ratio and the number of lexical items identified in the TOCS extension task for the deaf children ($r(27)$ = .576, p = .001) and the hearing children ($r(27)$ = .407, p = .028). The relationships between the social-tuning ratio and speechreading scores for both groups are depicted in Figure 5.

5.2.2.3 Group differences in the relationship between social-tuning ratio and speechreading scores

Deaf group only (training group contrast) – a multiple regression was calculated for the deaf group only to predict the number of lexical items identified in the TOCS extension task based on social-tuning ratio, the dummy coded variable for Group and the interaction between these two variables. The deaf participants' predicted number of lexical items identified in the TOCS extension task was equal to –24.68 + 109.57(Social-tuning ratio) + 29.21(Group) – 62.04(Interaction), where Group is coded as 0 = Maths, 1 = Speechreading. Social-tuning ratio was a significant predictor of speechreading scores ($p = .012$), but neither Group ($p = .072$) nor the interaction term ($p = .184$) were significant predictors. As there was no significant interaction between the speechreading and maths training groups, the deaf participants can be treated as a single group. When the interaction term is dropped from the model the number of lexical items identified on the speechreading task was equal to –11.65 + 72.94(Social-tuning ratio) + 6.37(Group). Social-tuning ratio ($p = .003$) was a significant predictor of speechreading scores, but the Group ($p = .195$) was not.

Deaf versus hearing groups – to test whether this relationship differed for deaf and hearing participants the number of lexical items identified in the TOCS extension task was predicted from the social-tuning ratio, Group (deaf = 0, hearing = 1) and the interaction between these variables. The participants' predicted number of lexical items identified in the TOCS extension task was equal to –9.97 + 77.28(Social-tuning ratio) + 14.67(Hearing status) – 50.90(Interaction). Hearing status ($p = .087$) was not a significant predictor of speechreading scores but both Social-tuning ratio ($p = .001$) and the interaction term ($p = .020$) were significant predictors of speechreading scores. This interaction confirms that the slope relating social-tuning score to TOCS speechreading was steeper in the deaf than the hearing group (see Figure 5).

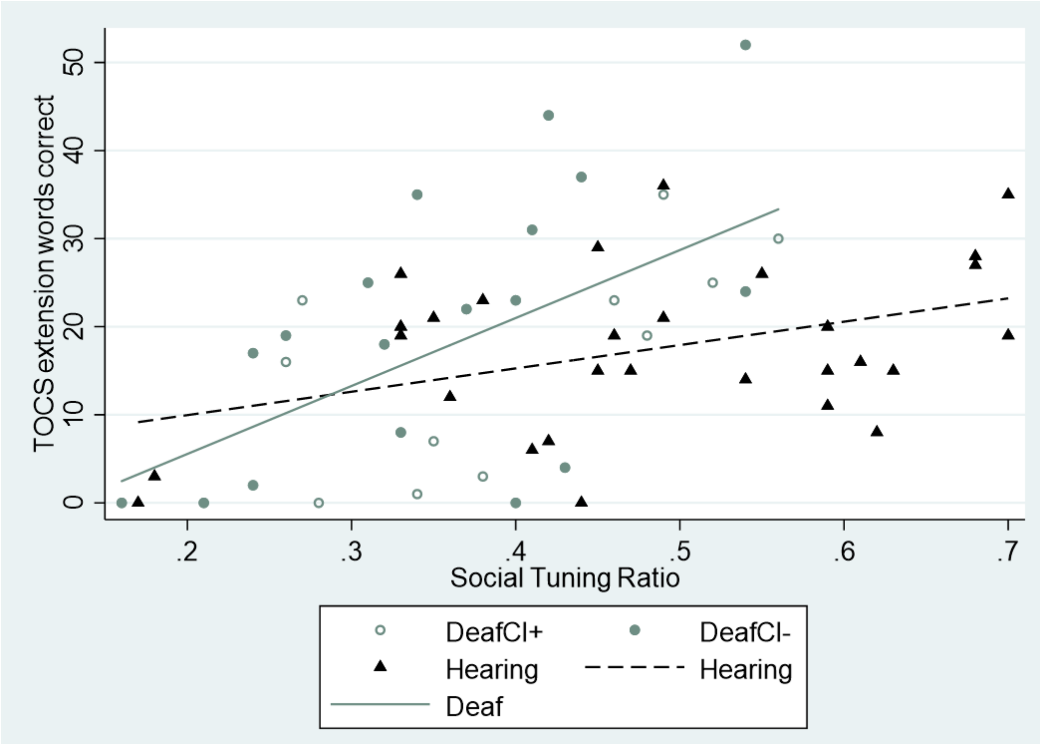

Figure 5. The number of words correctly identified in the TOCS Everyday Questions task plotted against 'social-tuning ratio' for both deaf and hearing participants. The relationship between these variables was significant for both groups. The slope for the deaf group (solid grey) was steeper than that for the hearing group (dashed black). To illustrate additional aspects of the heterogeneity of the deaf group, deaf children with CI are coded in hollow grey circles (n=11) and deaf children without CI are coded in solid grey circles (n= 18). Performance of deaf children with and without CI is not contrasted statistically due to small sample sizes.

In summary, the social-tuning ratio was higher in hearing than deaf children but correlated with speechreading scores in both groups, although this effect was stronger in the deaf than the hearing children.

5.2.2.4 Predicting reading scores using the social-tuning ratio

Hearing children used the social-tuning looking pattern more than deaf children. However, the relationship between this pattern and speechreading accuracy was stronger in deaf than in hearing children. We examined this pattern further in exploratory analyses. We reasoned that the social-tuning pattern may be related to conversational turn-taking skills, which in turn may be related to other language skills. In our battery we had two measures of reading, the YARC early word and single word reading subtests. A composite reading score was calculated by summing the Z-scores for each of the reading tests. There was a significant positive correlation between the social-tuning ratio and the composite reading score for the deaf children ($r(27) = .626$, $p < .001$) but not for the hearing children ($r(27) = .273$, $p = .152$).

In order to test whether this relationship differed for deaf and hearing participants a regression equation was calculated to predict the composite reading scores based on social-tuning ratio, the dummy-coded variable for hearing status and the interaction between these variables. The participants' predicted composite reading scores was equal to –4.06 + 10.92(Social-tuning ratio) + 2.39(Hearing status) – 7.49(Interaction), where Hearing status is coded as 0 = Deaf, 1 = Hearing. Social-tuning ratio ($p = .001$) was a significant predictor of reading scores but Hearing status ($p = .241$) and the interaction term were not ($p = .086$). When the interaction term is dropped from the model the composite reading score was equal to –2.26 + 6.08(Social-tuning ratio) – 0.68(Hearing status). In the deaf group alone, Social-tuning ratio ($p = .009$) was a significant predictor of composite reading scores, but the intervention group ($p = .177$) was not.

5.3 Discussion

Children born deaf must rely to a greater extent on visual input to access spoken language than hearing children. Despite this extensive difference in experience, here we show that young deaf and hearing children do not differ in speechreading accuracy or in the amount of time spent watching the mouth when watching silently spoken sentences (mouth-focus). In both groups mouth-focus correlated with the number of words children were able to identify from visual speech and the strength of this relationship did not differ between the deaf and hearing children. In addition, we found that both deaf and hearing children watched the eyes when the model was not speaking but watched the mouth during speech. This gaze pattern correlated with the children's speechreading performance, with a stronger relationship for the deaf than for the hearing children. These data provide insights into the mechanisms underlying speechreading success in deaf and hearing children. Each of these findings is discussed in detail below.

The absence of any difference in speechreading ability between deaf and hearing children is perhaps surprising. This finding is however consistent with previous research, showing no speechreading advantage in deaf over hearing children (Kyle et al., 2013; Kyle & Harris, 2006), despite such an advantage being observed in adults (Mohammed et al., 2006). In addition, deaf and hearing children did not differ in the amount of time spent watching the mouth during silent speech perception. To our knowledge this is the first time this issue has been addressed in children, deaf or hearing. Our findings are in line with previous research showing that hearing adults look at the mouth when auditory information is compromised during audiovisual speech perception in noise and when speechreading (Lansing & McConkie, 2003; Vatikiotis-Bateson et al., 1998).

For both deaf and hearing children, mouth-focus during the silent speechreading perception correlated positively with the number of words correctly identified. Furthermore, there was no difference between the deaf and hearing children in the strength of the relationship between mouth-focus and speechreading performance. This suggests that being born deaf, and relying on visual speech to access spoken language, does not affect how gaze behaviour relates to speechreading performance in early childhood. However, studies with adults suggest that developmental changes may be taking place that are yet to be documented. Two previous studies with hearing adults did not find a relationship between mouth-focus and speechreading (Lansing & McConkie, 2003; Wilson et al., 2016). Given that there appears to be a speechreading advantage for deaf adults over hearing adults (Bernstein et al., 2000; Mohammed et al., 2006; Pimperton et al., 2017), it is possible that gaze behaviour, and how this relates to speechreading ability, develops differently for deaf and hearing children at some point after the age range tested here, early childhood.

In addition to the predicted relationship between mouth-focus and speechreading performance, our analyses revealed a gaze pattern in which the children started each trial looking at the eyes before the speech started, shifted their gaze to the mouth during speech and returned to the eyes once the speech had finished. Both groups showed this social-tuning pattern, although the hearing children showed it more consistently than deaf children.

The extent to which children used the social-tuning pattern was positively correlated with the number of words correctly identified in the speechreading task. This was the case for both deaf and hearing children. These results suggest that those children who shift their attention between the eyes and the mouth when watching someone speak access more information from the spoken message than those who do not. Although causality is not clear from these correlational data, this pattern is consistent with data from hearing adults showing that gaze is task-dependent, suggesting that different areas of the face are more relevant for different types of information (Lansing & McConkie, 1999). Indeed, Lansing and McConkie (2003) found the same eyes-mouth-eyes gaze pattern identified here when hearing adults watched silently spoken sentences. They suggest that viewers are drawn to the eyes as a high contrast stimulus and because they have learned that the eyes express relevant social information, such as the talker's emotions and turn-taking. Turn-taking allows conversation to flow without speaking over another person and understanding turn-taking in conversation is important for language development (Rescorla, 1984). A reduction of gaze towards the eyes during speech has been shown to relate to impairment in language and social understanding in autistic individuals (Hanley et al., 2014). Therefore, the social-tuning pattern observed here may reflect the deaf and hearing children's understanding of turn-taking in conversation.

An alternative explanation for the social-tuning effect is that a moving mouth is a highly salient stimulus and therefore the participant's gaze may be drawn to it (Posner, 1980). However, if exogenous factors were the only explanation we would expect all participants to watch the mouth for the whole of the speech period. Instead, we observed substantial variation in the time the children spent looking at the mouth and the extent to which they used the social-tuning pattern. In addition, if eye gaze to the mouth was driven by visual salience alone, this would not explain the relationship we found between the social-tuning pattern and speechreading performance. Subsequent studies may be useful in directly testing hypotheses regarding the extent to which attention to the mouth during speech is driven by attention to movement.

The relationship between the social-tuning pattern and the number of words correctly identified in the speechreading task was stronger in the deaf than hearing children. As discussed above, the social-tuning pattern may reflect the deaf children's underlying language and communication skills. Although visual attention and turn-taking are important skills for language development in hearing children, they are particularly important for deaf children as they do not have full access to the social cues conveyed through prosody of the voice. We investigated the idea that the social-tuning pattern relates to other language-related skills in the deaf children more than hearing children by testing the relationship with reading proficiency. There was a significant relationship between the social-tuning pattern and single word reading scores in the deaf children but not in the hearing children. The interaction between hearing status and the social-tuning pattern was not significant but the effect size was relatively large ($\beta = -1.08$). Although these analyses were post-hoc and exploratory, the trend lends some support to the hypothesis that use of the social-tuning pattern is a stronger reflection of a deaf child's than a hearing child's broader language abilities. This difference should be investigated in future studies.

An important consideration for the current dataset is that the deaf participants were recruited from a large randomised controlled trial assessing the efficacy of a speechreading and reading intervention. Half the deaf participants received speechreading and reading training and half received maths training. The training groups did not differ in the extent to which they used the social-tuning pattern. However, children in the speechreading training group did spend significantly longer looking at the mouth than those in the maths training group. It is not possible to know whether or not gaze behaviour differed between the groups before training because no pre-training eyetracking data were collected. However, the mouth was emphasised during the speechreading training as an important location on the face. It is therefore possible that the speechreading training may have increased visual attention towards the mouth. Further studies should investigate this hypothesis directly.

5.3.1 Summary

The current findings provide insight into how young deaf and hearing children engage with a silently speaking face and how this relates to their speechreading ability. The results suggest that being born deaf, and therefore relying on visual information to access spoken language, does not change the way in which children access visual speech but does affect the extent to which they are able to benefit from employing specific gaze patterns. Although deaf children did not employ the social-tuning pattern more than hearing children, this pattern was more strongly related to speechreading skills in deaf than hearing children. This gaze pattern is suggested to reflect more general underlying language skills and showed a relationship to the deaf children's reading ability.

Given the relationship between speechreading and reading in both deaf and hearing children and how variable speechreading ability can be it is important to understand how children access visual speech information. Understanding the factors that relate to better speechreading skill in children may inform ways to improve speechreading. In turn, improving speechreading skill in deaf children may lead not only to better spoken language communication skills, but also to improved reading skills.

Visual speech information is one source of phonetic information for a deaf or hearing child to use to build robust phonological representations and is particularly important for deaf children. Looking at the mouth during speechreading is likely to provide a child with the maximum available phonetic information as this is the source of that information. Therefore, a child who looks more at the mouth during speechreading may be in a better position to build robust phonological representations than a child who does not. The following chapter will explore whether the relationship between speechreading and reading in deaf children can be explained by their phonological awareness skills.

6 The concurrent relationship between speechreading, phonological awareness and reading in deaf children

Chapter 4 reviewed evidence suggesting that speechreading ability is a strong concurrent and longitudinal predictor of reading ability in both deaf and hearing children. Similarly, phonological awareness correlates with reading ability in both deaf and hearing children (Harris et al., 2017b; Mayer & Trezek, 2014; Melby-Lervåg et al., 2012). As outlined in Chapter 2 speech is multimodal including both auditory and visual elements. Therefore, it is likely that phonological representations are multimodal too and that visual speech perception contributes to these representations. Several authors have suggested that deaf children may develop their phonological awareness through speechreading (Campbell & Dodd, 1980; Kyle, 2015; Kyle & Harris, 2011; Woll, 2012). This idea is discussed at length in Chapter 4.

In the current study we aimed to test the hypothesis that speechreading is related to phonological skills, which in turn are related to word reading (decoding skills). The model can be represented as a path model (speechreading -> phonological skills -> word reading). The causal steps procedure is the most common approach for assessing mediation (Baron & Kenny, 1986). This approach involves a series of three regression equations to establish the relationship between two variables (X and Y) and a potential mediating variable (M). The three equations involve: 1) regressing X onto Y;

2) regressing X onto M; 3) regressing both X and M onto Y. Regressing X onto Y establishes that a direct relationship exists between the two variables X and Y. Similarly, regressing X onto M establishes that these two variables are also related. Finally, regressing both X and M onto Y determines whether the effect of X on Y is reduced when M is included in the equation. If the variable M is mediating the relationship between X and Y it must be related to both X and Y and account for some, if not all, of the shared variance between X and Y.

However, one disadvantage with this traditional approach to mediation is that it involves regressions between single measures of particular constructs. Each measure will include some measurement error, which can lead to misleading results. Structural equation modelling (SEM) provides an opportunity to address this issue with latent variable path models (e.g. Fricke et al., 2013; Hulme et al., 2012; Melby-Lervåg, Lervåg, et al., 2012). Latent variables are factors that are assumed to exist but that are not directly measured. Instead, they are estimated using confirmatory factor analysis with multiple observed variables. Latent variables represent the shared variance from different measures, and therefore reflect a purer measure of the underlying construct (e.g. speechreading).

Another disadvantage with the traditional series of regression equations to assess mediation effects is that multiple relationships between factors cannot be represented in one equation. In contrast, SEM does allow for the investigation of simultaneous relationships between multiple factors, with a single factor being able to act as both a dependent variable and independent variable within the model. This means that the effect size for both the direct (i.e. from X to Y) and indirect (i.e. from X to Y via M) relationships between variables can be estimated. One other advantage of SEM is that extensive work has been done to establish robust methods for assessing the statistical significance of compound (mediating) paths.

SEM has several advantages over traditional mediation analyses, including modelling the relationship between the theoretical constructs of interest after controlling for measurement error and the ability to compare competing models. However, any analysis based on concurrent data cannot provide strong evidence for causal relationships. For any model with concurrent data there are many mathematically equivalent models that could fit the data but with different theoretical implications (see Thoemmes, 2015). Reversing the direction of regression equations will result in the same model fit and therefore these models cannot be differentiated from each other to inform about the direction of the relationship. Despite this, it is useful to establish the existence of a relationship between multiple factors and to describe the nature of those relationships. Therefore, SEM is a useful step to demonstrate a relationship between factors before using longitudinal or training studies to establish the direction of causality.

In summary, Chapter 4 established that one explanation for the relationship between speechreading and reading in young deaf children is that it is mediated by phonological awareness. Several studies have suggested that speechreading may contribute to phonological representations in young deaf children, which in turn may lead to better reading ability. Given the advantages of SEM for assessing mediation effects, in this chapter we used it to investigate whether the direct relationship between speechreading and reading is mediated by phonological awareness in 5-to-7-year-old deaf children. As with the other studies with deaf children in this thesis, the data were taken from the STAR_D project, which is described in detail in Appendix A.

6.1 Methods

For a detailed description of the participants and assessments included in the STAR_D project see Appendix A. The current study included all 66 STAR_D children (35 males), who were aged 6 years (*SD* = 7.8 months, range: 59-94 months) at the time of baseline data collection. The assessments used for the current model were as follows (see Appendix A for more details):

- **Speechreading:** The Test of Child Speechreading (TOCS; Kyle et al., 2013) Everyday Questions extension task. The score used was the number of words correctly identified from the silent videos.
- **Reading:** The YARC early word (EW) and single word (SW) reading subtests as well as the STAR_D in-house word-to-picture-matching task.
- **Phonological awareness:** The STAR_D onset- and rime-matching tasks.

6.2 Results

Descriptive statistics for the key measures included in the model for deaf children are shown in Table 2.

Table 2. Descriptive statistics for key measures in the model.

Task	M (*SD*)	Range	Cronbach's alpha
TOCS extension[a] (max = 62)	5.86 (*10.63*)	0-52	0.913
YARC EW reading[b] (max = 30)	11.00 (*10.54*)	0-30	0.975
YARC SW reading (max = 60)	7.02 (*10.15*)	0-43	0.967
STAR_D reading (max = 24)	11.76 (*6.90*)	1-24	0.915
Onset score (max = 12)	5.32 (*2.89*)	0-12	0.724
Rime score (max = 12)	5.18 (*2.69*)	1-12	0.695

[a]TOCS = Test of Child Speechreading [b]YARC = York Assessment of Reading Comprehension

Correlations between the measures of speechreading, phonological awareness and reading are shown in Table 3. All of the measures were highly correlated with one another ($p < .001$).

Table 3. Correlations between measures of speechreading, phonological awareness and reading.

Task	TOCS extension	YARC EW reading	YARC SW reading	STAR_D reading	Onset score	Rime score
TOCS extension[a]	-	.579	.631	.540	.451	.587
YARC EW reading[b]		-	.878	.923	.539	.502
YARC SW reading[b]			-	.841	.632	.611
STAR_D reading				-	.596	.529
Onset score					-	.506
Rime score						-

[a]TOCS = Test of Child Speechreading [b]YARC = York Assessment of Reading Comprehension. All of the measures were highly correlated with each other ($p < .001$).

The primary aim was to assess whether the direct relationship between speechreading and reading is mediated by phonological awareness. The analyses were conducted as a series of path models in Mplus (Version 8, Muthén & Muthén, 2017), using robust maximum likelihood estimators to account for some measures not being normally distributed. Any missing values were estimated using maximum likelihood estimators (default in Mplus). Effect sizes were estimated using standardised beta coefficients. The SEM analyses reported were carried out on the data collected at the baseline timepoint of the STAR_D project. That is, before the start of any training.

Therefore, unlike the eyetracking data presented in Chapter 5, Group does not need to be included as a factor in the analyses.

To assess the relationship between speechreading, phonological awareness and reading we constructed a latent variable mediation model with three constructs: Speechreading, Phonological Awareness and Reading. The latent variable for Speechreading was estimated by item parcelling every third word from the Test of Child Speechreading Everyday Questions extension task into three observed variables. For example, the TOCS observed variable 1 included words 1, 4, 7, 10, etc. and the TOCS observed variable 2 included words, 2, 5, 8, 11, etc. The three observed variables were then used as indicators for the speechreading latent variable. The purpose of item parcelling in this instance was to reduce the measurement error included in the regressions by creating a latent construct for speechreading based on three observed variables rather than regressing the TOCS extension task onto the latent constructs of phonological awareness and reading.

A latent variable for reading was estimated by performance on the YARC single word and early word reading subtests and the STAR_D reading test. A latent variable for phonological awareness was estimated from the STAR_D onset- and rime-matching tasks.

We used an iterative approach to construct the final model shown in Figure 6. In the first stage Speechreading was regressed onto Reading. There was a strong positive relationship between Speechreading and Reading ($\beta = 0.631$). In the second stage the Phonological Awareness latent variable was added as a mediating factor between Speechreading and Reading. Modification indices then indicated that correlation between STAR_D reading and YARC early word reading should be included in the model. This significantly improved the model fit (χ^2 difference = 12.43, df = 1, $p < .001$). This suggests that these two reading measures were correlated with each other in a way that was not accounted for via the Reading latent variable.

The error variance for single word reading was negative but not significant (a Heywood case). Therefore, for the final model the factor loading from the single word reading (YARC – SWRT) measure to the reading latent variable was fixed to 1. This did not change the fit of the model.

The final model showed full mediation, with the effect from Speechreading to Reading being entirely accounted for by the indirect pathway (speechreading – phonological awareness – reading). Dropping the non-significant direct path between Speechreading and Reading did not have a statistically significant effect on the model fit (χ^2 difference = 0.06, $p > .10$). Furthermore, the strength of the indirect pathway from Speechreading via Phonological Awareness to Reading did not change reliably when the non-significant direct pathway was dropped from the model. The indirect effect, via phonological awareness, was statistically reliable as assessed by a bias-corrected bootstrapped standard error (0.675, [95% CI 0.279, 5.509]).

Overall, this model provided an excellent fit to the data: $\chi^2(18, n = 66) = 20.539$, $p = .303$; comparative fit index = 0.996; Tucker-Lewis index = 0.993; RMSEA = 0.046 [95% CI 0.000, 0.123]; SRMR = 0.035. The model accounts for 74% of the variance in reading ability.

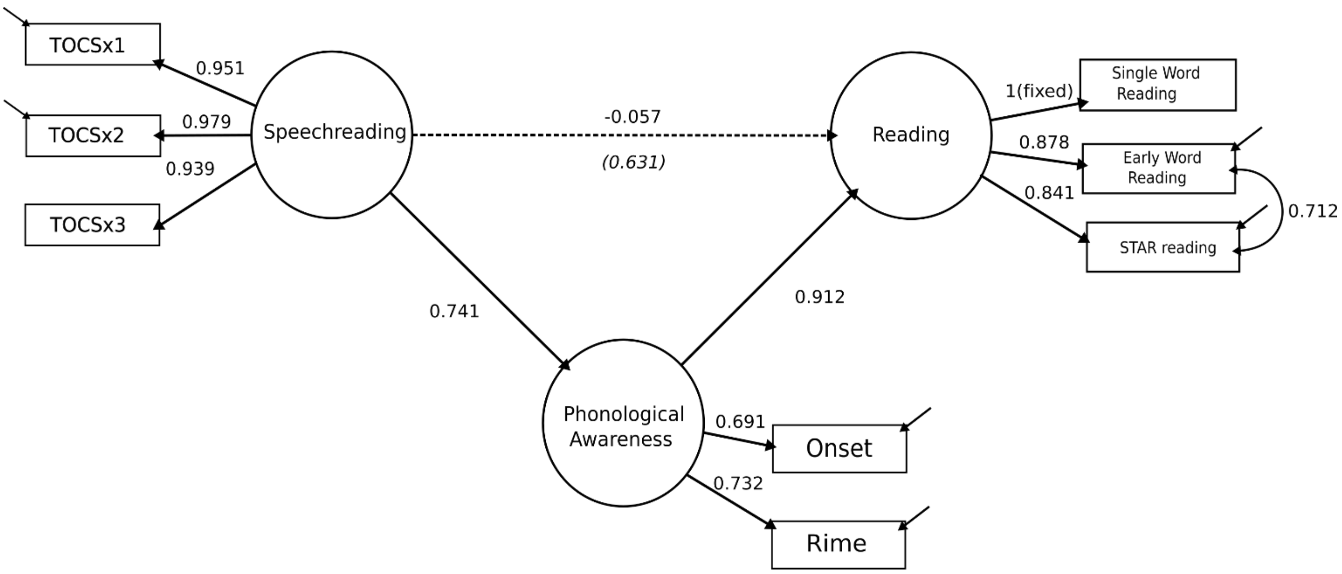

Figure 6. Path model showing the indirect relationship between speechreading and reading in young deaf children. Y-standardised regression coefficients are presented on the model. The Test of Child Speechreading was parcelled into three observed variables (TOCSx1, TOCSx2, TOCSx3) each of which contained a third of the items from this task. The dotted line indicates the direct path between speechreading and reading. The coefficient in brackets below the dotted line indicates the relationship before phonological awareness was included in the model. The coefficient above the dotted line indicates the non-significant path once phonological awareness was included in the model.

6.3 Discussion

The current analyses investigated whether the relationship between speechreading and reading ability in young deaf children is mediated by their phonological awareness skills. The study replicates the positive relationship between speechreading and reading in deaf children observed in previous studies (Harris et al., 2017b; Kyle et al., 2016; Kyle & Harris, 2006, 2011). In addition, this relationship between speechreading and reading was fully mediated by a measure of phonological awareness.

The model supports previous research showing that both speechreading and phonological awareness are related to reading ability in deaf children (e.g.

Campbell & Wright, 1988; Dyer, MacSweeney, Szczerbinski, Green, & Campbell, 2003; Harris & Beech, 1998; Harris et al., 2017; Kyle et al., 2016; Kyle & Harris, 2006, 2011). As described in Chapter 2, there is extensive debate about whether phonological awareness relates to reading ability in deaf individuals. Mayberry, del Giudice, & Lieberman (2011) suggested in their meta-analysis that phonological coding and awareness skills are related to reading ability in deaf children, but that they only explain a small amount of variance. However, Mayer & Trezek (2014) highlighted that the amount of variance in reading scores explained by phonological awareness was similar in deaf and hearing children. In addition, they pointed out that Mayberry et al.'s (2011) study included adults as well as children and that phonological awareness is known to be less predictive of reading ability in adults. The current results support the idea that in young deaf children phonological awareness skills do relate to reading ability ($R^2 = .736$).

Not only do these results support the idea that phonological awareness relates to reading ability, they also suggest that this may explain the relationship between speechreading and reading ability. This is consistent with suggestions from previous studies, that speechreading may allow for better specified phonological representations, which in turn improves a deaf child’s ability to map printed letters onto those representations in order to decode text (Kyle et al., 2016; Kyle & Harris, 2011; Mohammed et al., 2006). Mastering the alphabetic principle, that individual letters and groups of letters represent particular ‘sounds’ in spoken words (Byrne & Fielding-Barnsley, 1989), is an important part of learning to read. ‘Sounds’ is placed in inverted commas here because, as discussed in Chapter 2, phonological representations are likely to be multimodal rather than just representing sounds. In order to understand the link between letters and particular subcomponents in spoken language it is important to have robust representations of these subcomponents. This model is consistent with the idea that phonological representations are multimodal and that speechreading contributes to these representations in children with reduced access to auditory speech information (Kyle, 2015; Woll, 2012), which in turn improves reading.

The current study supports the idea that there is an indirect pathway between speechreading and reading via phonological awareness in young deaf children, as shown in Kyle's (2015) model. Kyle's (2015) model suggests that phonological awareness is established through both speechreading and reading and only then does it predict reading ability (see Kyle & Harris, 2010). Given that in the current model most children had started to learn to read, it is possible that the children’s phonological awareness skills had already been established to some extent via reading, speechreading and any auditory speech information to which the children had access. As these models are based on concurrent data after the children had started to read it is not possible to determine the direction of the relationship between these factors.

Mohammed et al. (2006) found that in deaf adults the relationship between speechreading and reading was maintained (r = .493) when controlling for phonological awareness scores, as measured by a picture-matching task based on onsets, rimes and individual phonemes. This suggests that although both speechreading and phonological awareness may relate to reading ability in deaf adults, there may be factors other than phonological awareness that explain the relationship between speechreading and reading.

One reason for the difference in results between the current study and Mohammed et al. (2006) may be that the relationships between speechreading, phonological awareness and reading change with age. Phonological awareness is less predictive of reading ability in older age-groups of hearing participants (Hogan et al., 2005; Wagner, Torgesen, Rashotte, Hecht, & Barker, 1997), possibly due to reduced variance in phonological awareness tasks, which reduces the potential shared variance with speechreading scores. However, it is also possible that the difference in results between the current study and Mohammed et al. (2006) is a result of methodological differences, as the tasks used to measure speechreading and reading differed substantially between the two studies. For example, in the current study, speechreading was measured with a free-response task rather than a multiple-choice task, which is likely to result in larger variance in scores. This means that there is greater potential for shared variance between speechreading, phonological awareness and reading scores.

One limitation of the current study is the relatively small sample size (66). This means that the model should be interpreted with caution. It should also be stressed that although the relationship between speechreading and reading is fully mediated by phonological awareness in the current model this does not preclude the possibility that other factors, such as vocabulary, may play a mediating role in this relationship (Rucker, Preacher, Tormala, & Petty, 2011). This idea will be explored further in the general discussion.

As the mediation model is based on concurrent data the direction of causality for the relationship between speechreading, phonological awareness and reading cannot be established. As outlined in the introduction, for a concurrent model there are many mathematically equivalent models that could fit the data but with different theoretical implications (see Thoemmes, 2015). Changing the direction of the arrows within the model would result in the same model fit but may have different effect sizes for the relationships between variables and different theoretical implications. However, Thoemmes (2015) demonstrated with simulations that the common assumption that the strongest indirect effect was the correct one is not true. Therefore, the direction of the relationship between the elements in this model cannot be determined from a concurrent dataset. Instead, either longitudinal data or experimental manipulation of speechreading is required. Despite the fact that causality cannot be determined from concurrent models, they provide a strong foundation for further investigation to establish the direction of the relationship between speechreading, phonological awareness and reading in deaf children.

In summary, the current model with young deaf children replicated the relationship between speechreading and reading ability. In addition, it established that this direct relationship was fully mediated by the children's scores on a phonological awareness task. The current model is consistent with the idea that visual speech perception may contribute to the development of robust phonological representations and provides a potential mechanism to explain the relationship between speechreading and reading in young deaf children.

7 The concurrent relationship between speechreading, phonological awareness and reading in hearing children

As introduced in Chapter 3 speech is by nature multimodal, involving visual as well as auditory information. It is well established that when visual speech is congruent with auditory speech it can enhance speech perception in adults (Lusk & Mitchel, 2016; Mitchel & Weiss, 2014; Sumby & Pollack, 1954), while incongruent visual information can disrupt it (McGurk & MacDonald, 1976). However, there is some debate about how useful visual speech information can be on its own in terms of accessing phonetic information. As described in Chapter 3 many speech sounds look the same on the lips as the sound is differentiated by articulators that cannot be seen, such as the vocal folds. For example, /m/, /p/ and /b/ all look the same. A category of sounds that look the same is referred to as a viseme. Some studies have suggested that given the proportion of visually indistinct speech sounds, viewers can access relatively little phonetic information from visual speech (Auer, Bernstein, Waldstein, & Tucker, 1997). However, others have shown that despite the overlap between different sounds in visual speech, it is still a rich source of phonetic information (Bernstein et al., 2000; Buchwald, Winters, & Pisoni, 2009; Sell & Kaschak, 2009; Soto-Faraco et al., 2007).

Sell and Kaschak (2009) trained hearing adults with continuous speech streams of an artificial language either with auditory-only information, visual-only information or audiovisual information. They found that adults were able to identify test words from the artificial language that were presented in the auditory modality only, regardless of what sort of training they had received. This suggests that visual speech information can be used to segment a continuous speech stream and that information can be transferred into the auditory modality. This has implications for the role of visual information in infants learning their language as it may help them to identify words.

In hearing adults, visual speech information can enhance auditory speech perception in noise even when the two signals are not simultaneous. Buchwald, Winters and Pisoni (2009) used silent videos to visually prime degraded auditory words. They found that the hearing adults in this study were faster to identify the degraded auditory words when preceded by a matching visual-speech target. In addition, the incorrect responses that the participants provided were often correct regarding the place of articulation shown in the visual prime. These results suggest that hearing adults are able to access phonetic information from the visual-only stimulus and can map this onto the auditory stimulus. This supports the idea that although many speech sounds overlap visually, visual speech can still provide relevant phonetic information to the perceiver. This is further supported by findings that hearing adults can discriminate between two spoken languages if they have knowledge of at least one of those languages (Soto-Faraco et al., 2007) based on speechread information only.

Research with hearing adults supports the idea that visual speech is a rich source of phonetic information despite the overlap in visual articulation of many speech sounds. The cross-modal transfer of information from the visual to the auditory modality supports the idea that phonological representations are multimodal and that both visual and auditory information contribute to those representations.

In hearing infants, speech perception abilities have been shown to be related to later language development. Kuhl, Conboy, Padden, Nelson and Pruitt (2005) showed that 7-month-olds' ability to discriminate between auditory presentations of native phonetic contrasts (such as /ta/ and /pa/) predicted development in several different language abilities such as the number of words produced, sentence complexity and mean length utterance at 18 and 24 months old. They suggested that this relationship between speech perception skills and language development can be explained by phonetic discrimination ability aiding detection of statistical regularities in the speech stream, segmentation of speech and word identification. Not only does auditory speech perception relate to language development but other studies have also suggested that visual speech perception influences word-form recognition (Weatherhead & White, 2017) and vocabulary development in infants and young children (Altvater-Mackensen & Grossmann, 2015; Erdener & Burnham, 2018; Jerger et al., 2018). Altvater-Mackensen and Grossmann (2015) presented 6-month-olds with auditory and visually presented vowels that either matched or mismatched. They found that the infants' ability to identify mismatched phonetic information across modalities was correlated with their later vocabulary skills. This suggests that visual speech perception plays a role in language development in hearing children.

Given that visual speech perception can influence auditory speech perception, it is likely that visual speech information also provides hearing infants with important information for segmenting speech and learning the phonetic boundaries of their native language. Hearing infants are sensitive to visual speech information from a young age. Both Kuhl and Meltzoff (1982) and Patterson and Werker (1999) found that 4.5 month old infants were able to recognise which of two speaking faces matched an auditory speech sound, which has since been shown in infants as young as 2 months old (Patterson & Werker, 2003). These studies suggest that young infants may have an integrated representation of auditory and visual speech information. Being able to detect the correspondence between auditory and visual speech information is likely to help infants to segment speech and learn phonetic categories as they have more than one source of information to base this on. Teinonen, Aslin, Alku and Csibra (2008) presented 6-month-old hearing infants with an auditory stimulus along the /ba/-/da/ continuum with a unimodal frequency distribution centred on the category boundary. One group of infants saw an accompanying visually-presented /ba/ and /da/ corresponding to the categories (two-category visual group) and the other group saw either a /ba/ or a /da/ for the whole continuum (single-category visual group). When tested on tokens either side of the category boundary with auditory information only, the two-category visual group were able to discriminate the items, but the one-category visual group were not. These results suggest that not only does visual speech information influence auditory speech perception but it can also influence learning of perceptual categories in 6-month-old hearing infants. In addition to discriminating phonetic information using both auditory and visual speech information, 4-month-old infants can also discriminate between native and non-native phonetic contrasts from visual-only information (Weikum et al., 2007). Hearing children can also use speechread information to both identify and discriminate phonemes (Jerger et al., 2018). This suggests that visual speech on its own can be used to access phonetic information to aid language development in young hearing infants and children.

As described above, being able to discriminate between phonemes in their native language will help infants to detect regularities in the language they are learning, to segment speech and thus to identify words. However, although infants are sensitive to visual speech information, its influence on speech perception still takes some time to become adult-like with children being less sensitive to the McGurk effect than adults as described in Chapter 3 (Massaro, Thompson, Barron, & Laren, 1986; McGurk & MacDonald, 1976; Sekiyama & Burnham, 2008). In addition, the attenuation of auditory event-related potentials by visual speech information is still developing at 12 years of age although different aspects mature at different rates (Knowland, Mercure, Karmiloff-Smith, Dick, & Thomas, 2014). Knowland et al. (2014) suggested that although children may be able to access phonetic information from visual speech cues, it may take longer to use visual speech information to make predictions about the lexical content of the auditory speech stream in an adult-like way. This suggests that although the influence of visual speech information continues to develop throughout childhood, children may still use visual speech information to build robust phonological representations from an early stage.

Speechreading ability has been found to be associated with phonological awareness ability in hearing children with and without developmental language disorder (Heikkilä, Lonka, Ahola, Meronen, & Tiippana, 2016). In addition, speechreading skill is associated with phonological awareness ability in hearing adults with developmental dyslexia (Mohammed et al., 2006). These correlational studies suggest that visual speech perception may contribute to the development of multimodal phonological representations, which was tested by Heikkilä et al. (2018). They trained 7-to-10-year-old children with developmental language disorder, who tend to be less influenced by visual speech information than their typically developing peers (Heikkilä et al., 2016; Knowland et al., 2016), with either auditory-only speech perception or on audiovisual speech perception. They found that the audiovisual speech perception training led to improvement on a non-word repetition task, which was used as a measure of phonological representations, whereas the auditory-only training did not. This study highlights the multimodal nature of speech and phonological representations emphasising the role of visual speech perception in the development of phonological representations in hearing children.

In summary, given the role of visual speech information in audiovisual speech perception, it is likely that visual information contributes to phonological representations in hearing children even though they have full access to auditory speech information. Indeed, infants as young as 2 months old are sensitive to the correspondence between auditory and visual speech information and visual information has been shown to influence phonetic discrimination, which in turn predicts language development. Therefore, it is perhaps not surprising that speechreading ability relates to reading ability in hearing children (Kyle et al., 2016; Kyle & Harris, 2010) as well as deaf children. Chapter 6 described a series of path models suggesting that in young deaf children the relationship between speechreading and reading is fully mediated by phonological awareness skills. Therefore, in the current study we aimed to investigate whether the relationship between speechreading and reading in hearing children is mediated by phonological awareness, as it is in deaf children. We tested young hearing children (aged 5-8 years) on measures of speechreading, phonological awareness and reading similar to those used in Chapter 6 with deaf children. Some of the tests that had been used with deaf children in Chapter 6 were changed to be more appropriate for use with hearing children.

7.1 Methods

7.1.1 Participants

A group of 138 5-8-year-old hearing children (79 males) were recruited from two mainstream schools in Cambridgeshire and one mainstream school in London. Two children were excluded, one because they did not speak English, and the second because they withdrew their participation. Of the remaining children, 91 children were monolingual English speakers, 32 spoke an additional language at home but had learnt English from birth and the remaining 13 children had been learning English for an average of 3 years (*SD* = 1.2 years; range: 1-5 years), as reported by their parents. Despite this, these 13 children had an average T-score of 51 on the British Ability Scales (BAS) Word Definitions subtest (group range: 38-64). They were therefore included in the study. The hearing children had a mean age of 6 years and 5 months (*SD* = 8.5 months).

7.1.2 Measures

7.1.2.1 Vocabulary screening measure

The BAS Word Definitions subtest was used to screen children’s vocabulary levels as described above. In this task, words are read to a child and they are asked to give a definition of the word.

7.1.2.2 Speechreading

As with the deaf children, speechreading ability was assessed in these hearing children using the TOCS Everyday Questions extension task.

7.1.2.3 Reading

Reading was assessed using the YARC early word and single word reading subtests, both of which were used as measures of reading in the deaf children in Chapter 6. However, the STAR_D reading task that was used in Chapter 6 was not included in the reading test battery with the hearing children. This is because the hearing children were all able to give a verbal response to the reading tasks and multiple-choice tasks are less sensitive than free-response tasks.

7.1.2.4 Phonological awareness

Phonological awareness was assessed using the YARC sound deletion subtest (Hulme et al., 2009) as this task is known to be highly predictive of reading ability in hearing children (e.g. Hulme et al., 2012; Thompson et al., 2015). In this task the children were asked to repeat words with particular sounds removed, for example "Can you say frog? … Can you say it again without the /r/?". The task involves deleting whole segments of compound words (e.g. 'saw' from 'seesaw'), initial consonants, final consonants and internal consonants from consonant clusters. Each deletion type is introduced with one or two practice trials for which feedback is given and children can score a total of 12 points. Again, this task differed from the phonological awareness task used with the deaf children in Chapter 6 in order to increase measurement sensitivity and because there was no requirement for a non-verbal response task.

7.2 Results

Descriptive statistics for the vocabulary screening measure (BAS Word Definitions) and the raw scores on key measures included in the model for hearing children are shown in Table 4.

Table 4. Means (standard deviations) and reliabilities for the measures used in the study

Task	M (*SD*)	Range	Cronbach's alpha
TOCS extension[a] (max = 62)	16.95 (*10.10*)	0-47	0.92
YARC EW reading[b] (max = 30)	24.63 (*7.56*)	0-30	0.96
YARC SW reading[b] (max = 60)	25.92 (*10.69*)	0-56	0.97
Phoneme deletion (max = 12)	7.96 (*2.75*)	0-12	0.79
BAS Word Definitions[c] (max = 27)	10.94 (*4.01*)	3-18	0.84

[a]TOCS = Test of Child Speechreading [b]YARC = York Assessment of Reading Comprehension [c]BAS = British Ability Scales.

Correlations between the measures of speechreading, phonological awareness, reading and the vocabulary screening measure are shown in Table 5. All correlations were significant ($p < .001$).

Table 5. Correlations between measures of speechreading, phonological awareness, reading and the vocabulary screening measure

Task	TOCS extension	YARC EW reading	YARC SW reading	Phoneme deletion	BAS Word definitions
TOCS extension[a]	-	.494	.564	.471	.364
YARC EW reading[b]		-	.814	.771	.485
YARC SW reading[b]			-	.759	.539
Phoneme deletion				-	.452
BAS Word Definitions[c]					-

[a]TOCS = Test of Child Speechreading [b]YARC = York Assessment of Reading Comprehension [c]BAS = British Ability Scales. All correlations were significant ($p < .001$).

The primary aim was to assess whether the relationship between speechreading and reading is mediated by phonological awareness. As with the deaf children, a series of path models were conducted in Mplus (Version 8). As before, robust maximum likelihood estimators were used to account for any measures not being normally distributed. In addition, the small number of missing values were dealt with full information maximum likelihood estimators (default in Mplus). Effect sizes were estimated using standardised beta coefficients.

To assess the relationship between speechreading, phonological awareness and reading we constructed a latent variable mediation model with three constructs: Speechreading, Phonological Awareness and Reading. The latent variable for Speechreading was estimated using item parcelling of the TOCS extension subtest as with the deaf model presented in Chapter 6. A latent variable for reading was estimated by performance on the YARC single word and early word reading subtests. A latent variable for Phonological Awareness was defined by only one indicator, the YARC sound deletion subtest with a reliability of 0.79. There were not sufficient items (12) in the sound deletion task to conduct item parcelling. Therefore, the error variance was set to 1.58, which was variance (7.568) minus variance multiplied by reliability of the measure (0.79). This approach was used to account for measurement error in the phonological awareness scores.

We used an iterative approach to construct the final model shown in Figure 7. In the first stage Speechreading was regressed onto Reading. There was a strong positive relationship between Speechreading and Reading ($\beta = 0.610$). In the second stage the Phonological Awareness latent variable was added as a mediating factor between Speechreading and Reading. The final model showed a full mediation, with the effect from Speechreading to Reading being entirely accounted for by the indirect pathway (Speechreading – Phonological Awareness – Reading). The indirect effect, via phonological awareness, was statistically reliable as assessed by a bias-corrected bootstrapped standard error (0.505 [95% CI 0.341, 0.708]). Dropping the non-significant direct path between speechreading and reading did not have a statistically significant effect on the model fit (χ^2 difference = 0.81, $p > .10$). Furthermore, the strength of the indirect pathway from speechreading via phonological awareness to reading did not change reliably when the non-significant direct pathway was dropped from the model.

Overall, this model provided an excellent fit to the data: $\chi^2(8, n = 136) = 7.907$, $p = .443$; comparative fit index = 1; Tucker-Lewis index = 1, RMSEA < .001 [95% CI 0.000, 0.100]; SRMR = 0.026. The model accounts for 82% of the variance in reading ability.

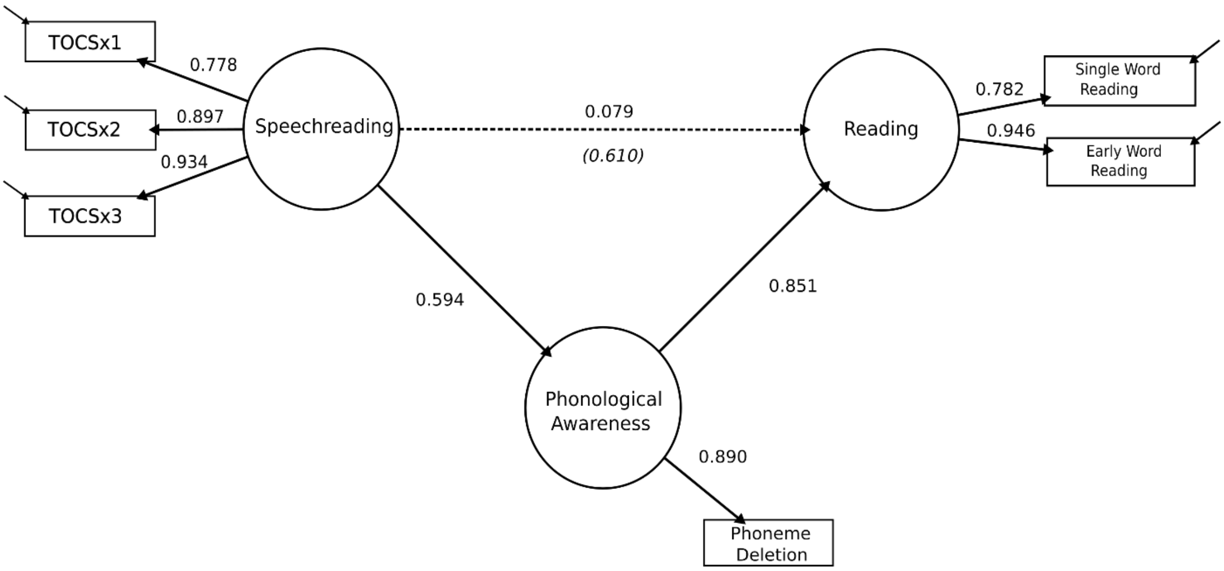

Figure 7. Path model showing the indirect relationship between speechreading and reading in hearing children. Y-standardised regression coefficients are presented on the model. The Test of Child Speechreading was parcelled into three observed variables (TOCSx1, TOCSx2, TOCSx3) each of which contained a third of the items from this task. The error variance for phoneme deletion was set to 1.58 to account for measurement error. The dotted line indicates the direct path between speechreading and reading. The coefficient in brackets below the dotted line indicates the relationship before phonological awareness was included in the model. The coefficient above the dotted line indicates the non-significant path once phonological awareness was included in the model.

7.3 Discussion

The current study examined whether the relationship between speechreading and reading ability in young hearing children is mediated by phonological awareness skills. The model supports the idea that in young hearing children, speechreading and reading are strongly related to each other. In addition, this direct relationship between speechreading and reading was fully mediated by phonological awareness.

The relationship between speechreading and reading is consistent with previous findings showing that speechreading relates to reading accuracy in hearing children (Kyle et al., 2016; Kyle & Harris, 2010) not just in deaf children. Direct comparisons between the models with deaf and hearing children cannot be made as different tasks were used to measure reading and phonological awareness for each of these models. However, the similarity between the models is striking, given that hearing children have full access to auditory speech information and are therefore likely to rely less on visual speech information than deaf children. Therefore, although visual speech perception has been shown to relate to language development (Weatherhead & White, 2017) and phonetic discrimination (Erdener & Burnham, 2013; Weikum et al., 2007) in hearing children, it might be expected to relate to phonological awareness and reading ability to a lesser extent than in deaf children. However, the current studies suggest that the relationship between speechreading, phonological awareness and reading is very similar in deaf and hearing children.

One of the limitations with the model with deaf children in Chapter 6 was that it was based on only 66 participants, which is a relatively small sample for structural equation modelling. However, the model with hearing children is based on a much larger sample and shows a very similar pattern of relationship between speechreading, phonological awareness and reading to the model with deaf children. This lends support to the model with deaf children.

The model with hearing children not only supports the work with deaf children but is also consistent with the idea that visual speech information contributes to multimodal phonological representations in hearing children (Kyle et al., 2016; Mohammed et al., 2006). Visual and auditory speech information provide redundant information, allowing for more robust, multimodal representations of phonological information. As with deaf children, hearing children who have more robust representations of the subcomponents of spoken language are likely to find it easier to master the alphabetic principle and therefore to learn to read. The current model with hearing children suggests that although deaf children do rely on speechreading to a greater extent than hearing children to access spoken language this does not account for the full mediation shown in young deaf children in Chapter 6.

The relationship between speechreading and phonological awareness observed in the current model is consistent with previous findings in populations who tend to have weaker phonological skills. Hearing children with developmental language disorder and hearing adults with developmental dyslexia both show a relationship between speechreading and phonological awareness ability (Heikkilä et al., 2016; Mohammed et al., 2006). Similarly, studies with typically developing hearing children have shown a relationship between speechreading and phonological awareness (Kyle & Harris, 2011; Lyxell & Holmberg, 2000). However, hearing adults do not show a relationship between speechreading and phonological awareness or speechreading and reading (Mohammed et al., 2006). In addition, a recent study by Harris, Terlektsi, & Kyle (2017) found that speechreading was not related to reading ability or phonological awareness ability in 6-year-old hearing children either concurrently or as a longitudinal predictor. One explanation for these studies finding no relationship between speechreading and phonological awareness is that there was low variability in phonological awareness scores in both the hearing adults and children. Harris et al. (2017) noted that their phonological awareness task was aimed at the deaf children in their study and many of the hearing children tested were at ceiling in this task making it hard to observe a relationship between phonological awareness and speechreading ability. This issue was avoided in the current model with hearing children by having a different measure of phonological awareness for the deaf and hearing children, meaning that the two models are not directly comparable but that there were no ceiling effects for the phonological awareness task in hearing children.

Although the current study supports studies showing a relationship between speechreading and phonological awareness a recent study by Snowling, Lervåg, Nash and Hulme (2018) called into question what the contribution of speech perception to phonological representations might be. The authors used structural equation modelling to investigate the relationship between auditory speech perception and reading ability in children at risk of developmental dyslexia and in typically developing children. They found that auditory speech perception, as measured by a categorical perception task, related concurrently to phoneme awareness skills, and that both of these skills related to later reading ability, which is in line with the current findings. Together these results suggest that speech perception skills, whether visual or auditory, relate to phonological awareness and in turn reading. However, Snowling et al.'s (2018) model included a longitudinal mediation analysis, which showed that the direct relationship between auditory speech perception at age 5 ½ and reading ability at age 6 ½ was not mediated by phoneme awareness skills. This challenges the idea that speech perception abilities play a causal role in the development of reading skills through phoneme awareness skills. However, Snowling et al. (2018) noted that the categorical perception task used taps several other skills beyond speech perception, such as attention and decision-making skills. This may explain to some extent the relationship observed between categorical perception and reading that was separable from the effect of phoneme awareness. Further studies are needed to investigate the nature of the relationship between speech perception, both auditory and visual, tested in a number of different ways, and reading development. It may also be that speech perception is important earlier in development, although this would be hard to measure in younger children.

Although the models with deaf and hearing children are consistent with the idea that speechreading contributes to multimodal phonological representations, explaining the relationship between speechreading and reading, it is possible that the direction of the relationship is the other way round. Particularly for hearing children, who have full access to auditory speech information, it may be that phonological awareness has a causal effect on both speechreading and reading rather than mediating the relationship between them. They may build their phonological representations primarily through auditory speech perception and better phonological representations may allow a child to map them onto orthography and visual speech resulting in better reading and speechreading respectively. However, for deaf children speechreading is a key source of information about the structure of spoken language. Therefore, although deaf children may be able to develop their phonological representations through reading to some extent (Kyle, 2015), speechread information is likely to contribute to the development of phonological representations (Kyle, 2015).

Hearing children may be able to establish phonological representations primarily through auditory speech perception and then use these representations to help their speechreading. However, many studies indicate that visual speech information can influence speech perception in both hearing adults and infants (Lusk & Mitchel, 2016; Mitchel & Weiss, 2010; Sumby & Pollack, 1954; Teinonen et al., 2008). In addition, in order for hearing children to be able to map from auditory speech representations to speechreading they must have multimodal phonological representations to make this cross-modal transfer of information. Therefore, although it is possible that phonological awareness is acting causally on both speechreading and reading as opposed to being a mediator of the relationship between speechreading and reading in hearing children, it is still likely that visual speech perception contributes to establishing robust multimodal phonological representations.

It is likely that the relationships between the three elements of the model, speechreading, phonological awareness and reading, are reciprocal and that the direction of these relationships may change over the course of development. However, as described in Chapter 6 the mediation models with deaf and hearing children are based on concurrent data and therefore there are many other mathematically equivalent models that could fit the data but with different theoretical implications (see Thoemmes, 2015). In order to differentiate between equivalent models, either longitudinal data or experimental manipulation of speechreading is required. However, the concurrent models provide a strong foundation for this kind of further investigation to establish the direction of the relationship between speechreading, phonological awareness and reading in deaf and hearing children.

8 Speechreading training in hearing children

As discussed in Chapters 3 and 7 visual speech perception is important for hearing as well as for deaf individuals, boosting auditory speech perception in noise (Lusk & Mitchel, 2016; Mitchel & Weiss, 2014; Sumby & Pollack, 1954) and promoting phonetic discrimination and language development in hearing infants (Altvater-Mackensen & Grossmann, 2015; Erdener & Burnham, 2018; Jerger et al., 2018; Teinonen et al., 2008). It is well established that phonological awareness relates to reading ability in hearing children (Caravolas et al., 2012; Castles & Coltheart, 2004; Hatcher et al., 2004; Hulme et al., 2002; Snowling & Hulme, 2005). In addition, speechreading ability is related to both reading ability and phonological awareness in hearing children (Heikkilä et al., 2016; Kyle et al., 2016; Kyle & Harris, 2010). Chapters 2 and 4 suggested that visual speech perception may contribute to the development of multimodal phonological representations in hearing children as well as deaf children because of its overlap with auditory speech perception. The mediation model presented in Chapter 7 showed that speechreading and reading are related to each other and that this relationship is mediated by phonological awareness in young hearing children. This supports the idea that visual speech perception may play a role in the development of multimodal phonological representations in hearing children, which in turn may support reading development. However, as the model was based on concurrent data it cannot demonstrate whether there is any causal relationship between speechreading and reading.

Phonological awareness is known to be an important predictor of reading development in hearing children (see Chapter 2). Those with poorer phonological awareness, such as those with developmental dyslexia or developmental language disorder (DLD), tend to have difficulties with reading (Leonard, 2014; Snowling, 2000). Given the importance of phonological awareness in hearing children's reading development it is of interest to understand how phonological representations and awareness develop. As described above, visual speech perception may contribute to the development of multimodal phonological representations. Individuals with developmental dyslexia and DLD tend to have poorer visual speech perception and speechreading skills than their typically developing peers (Heikkilä et al., 2016; Knowland et al., 2016; Mohammed et al., 2006). Poorer speechreading ability in these groups may be a consequence of them having poorer phonological skills. However, it is also possible that poorer visual speech perception ability gives rise to less robust phonological representations in these groups. It could be that improving speechreading ability in hearing children with poorer phonological skills would provide them with another route, in addition to auditory information, into the structure of spoken language and so allow them to develop more robust phonological representations. The current study aimed to determine whether visual speech information contributes to phonological awareness ability in hearing children.

Heikkilä et al. (2018) suggested that visual speech perception does play a causal role in the development of phonological representations in hearing children with DLD. They trained children with DLD to match words to letters, pictures and syllables. Half of the children were trained with auditory-only speech and the other half with auditory-visual speech. The children were trained for six weeks, five days a week for 10-15 minutes a day. Heikkilä et al. (2018) found that the children trained in the auditory-visual group, but not the auditory-only group, improved in a non-word-repetition test, which was used as a measure of phonological representations. They suggested that visual speech cues aided the development of phonological skills as being able to see the articulatory gestures may help children to disambiguate speech sounds. This supports the idea that visual speech information contributes to phonological representations.

One way to assess the causal contribution of visual speech information to the development of phonological representations and awareness is to train speechreading. Some researchers have claimed that speechreading is a 'hard-wired' skill that cannot be trained (e.g. Summerfield, 1992). However, as described in Chapter 3 speechreading ability is known to improve with age (Kyle et al., 2013) and deaf adults tend to be better at speechreading than hearing adults (Bernstein et al., 2000; Mohammed et al., 2006; Pimperton et al., 2017) despite no differences in speechreading skill between deaf and hearing children. This suggests that experience with speechreading can improve this skill. In addition, many studies have shown improvements, albeit moderate, in speechreading skill in deaf and hearing adults following training (Bernstein et al., 2001; Blumsack et al., 2007; Bothe, 2007; Lonka, 1995; Walden et al., 1977). More recently, work with 8-to-10-year-old hearing children has shown that a single session of training with Cued Speech and speechreading leads to improvements in Cued Speech perception (Rees, Fitzpatrick, Foulkes, Peterson, & Newton, 2017). This suggests that speechreading may be trainable in hearing children.

The STAR_D project (Appendix A) was a randomised controlled trial to investigate whether speechreading training would lead to improvements in phonological awareness and consequently reading ability in young deaf children. The speechreading intervention group showed greater gains in speechreading on the Test of Child Speechreading Everyday Questions subtest than the maths control group. The speechreading training did not improve phonological awareness (onset and rime judgement) or single word reading. However, the training group did show greater improvements on speech production, which was used as a proxy for their phonological representations. Therefore, although no improvements were seen on an explicit measure of phonological awareness and reading as a result of the training, this study demonstrated that speechreading can be trained in young deaf children and that this may relate to phonological representations.

There are several possible reasons why no improvement was observed on the phonological awareness task as a result of speechreading training within the STAR_D project. One reason may be that the phonological awareness assessment used was multiple-choice, with 3 options per trial, making this a less sensitive measure than a free-response task. In addition, the sample size was quite small and therefore the study had low statistical power.

Given the similarity of SEM analyses of the concurrent data between deaf and hearing children in Chapters 6 and 7 it is possible that training speechreading in hearing children may result in similar outcomes to training speechreading in deaf children. Improving speechreading ability may improve hearing children's access to the phonetic information in speech, allowing them to build more robust multimodal phonological representations, which may in turn help those with poorer reading skills. Therefore, the aim of the current study was to investigate the contribution of visual speech information to phonological awareness skills and reading ability in young hearing children. Specifically, this was a proof-of-principle study that aimed to test whether training speechreading would lead to improvement on phonological awareness tasks using trained and untrained words and if so, whether there was a knock-on effect on reading ability. The STAR_D training programme was adapted for use with 4-5-year-old hearing children based on the deaf children's performance and teacher feedback from the STAR_D project.

We tested 92 hearing children aged 4-5 years old on measures of speechreading, reading and phonological awareness. The children were then randomly assigned to two groups, one of which completed 3 weeks of speechreading training games for 10 minutes a day, 5 days a week, with the other group acting as the business-as-usual (BAU) controls. The controls did not receive any training as part of the project but all Reception children received their standard 30 minutes of phonics lessons in school every day. Both groups were tested again immediately after the training and then followed up three months later. The speechreading, phonological awareness and reading tests were comprised of a set of words used in the training and a matched set of untrained words.

The current study investigated whether speechreading training with hearing children can improve speechreading, phonological awareness and reading:

1) In relation to speechreading we assessed three hypotheses:

i. The speechreading training group will perform better than the control group on post-tests of single word speechreading when controlling for baseline scores.

ii. Within the speechreading training group, the children will be better at speechreading trained items than untrained items.
iii. The speechreading training group will perform better than the control group on a general measure of sentence speechreading (new models and stimuli).

2) In relation to phonological awareness we assessed two hypotheses:

i. The speechreading training group will perform better than the BAU control group on post-tests of phonological awareness when controlling for baseline scores.
ii. Within the speechreading training group, the children will be better at phonological awareness tasks with trained items than untrained items.

3) For reading, we assessed two hypotheses:

i. The speechreading training group will perform better than the control group on post-tests of single word reading when controlling for baseline scores.
ii. Within the speechreading training group, the children will be better at reading trained items than untrained items.

8.1 Methods

8.1.1 Design

The design for this study was pre-registered on the Open Science Framework (https://osf.io/wyc84/register/565fb3678c5e4a66b5582f67). This study was a randomised experiment with hearing children aged 4-5 years old. Children were assessed at pre-test (T1) on a battery of assessments of speechreading, phonological awareness and reading. They were then randomised into those who undertook the speechreading intervention and a business-as-usual control group. They were then assessed again immediately after the intervention period (T2). After the T2 assessment it was decided to follow up the children after 3 months (T3) to assess whether the intervention effects were maintained. The T3 assessments were not included in the pre-registration and so the T3 analyses are reported here as exploratory. The intervention was run by the experimenter and therefore was not blinded.

8.1.2 Participants

Ninety-two 4-to-5-year-old hearing children were recruited from mainstream schools in Cambridgeshire and London. The CONSORT diagram in Figure 8 shows the flow of participants through the trial. Two children were excluded because they were unresponsive in baseline tasks and four were excluded as they had a vocabulary score more than two standard deviations away from the mean of the group (M = 37.26, SD = 4.73, cut-off was 27.80). The remaining 86 children (38 females) had a mean age of 4 years and 11 months (SD = 3.7 months; range: 52-65 months) at baseline. The children participated in this study in the second term of their Reception year at school. Pilot work indicated that children at the beginning of their first term of school were not able to complete any of the assessment tasks but children in the first term of Year 1 (second year at school) were already at ceiling on many of the phonological awareness tasks. By the end of their first term in reception at school, pilot children were able to attempt most of the tasks. Therefore, conducting the intervention in the second term of school was considered appropriate.

The children were randomised by Professor Charles Hulme using stratified randomisation within classes and schools in Stata (Version 15.1; StataCorp. 2017). There were 43 children (18 female) in the intervention group and 43 (20 female) in the BAU control group and there were no differences between the groups in age or performance on any of the three phonological awareness tasks at baseline.

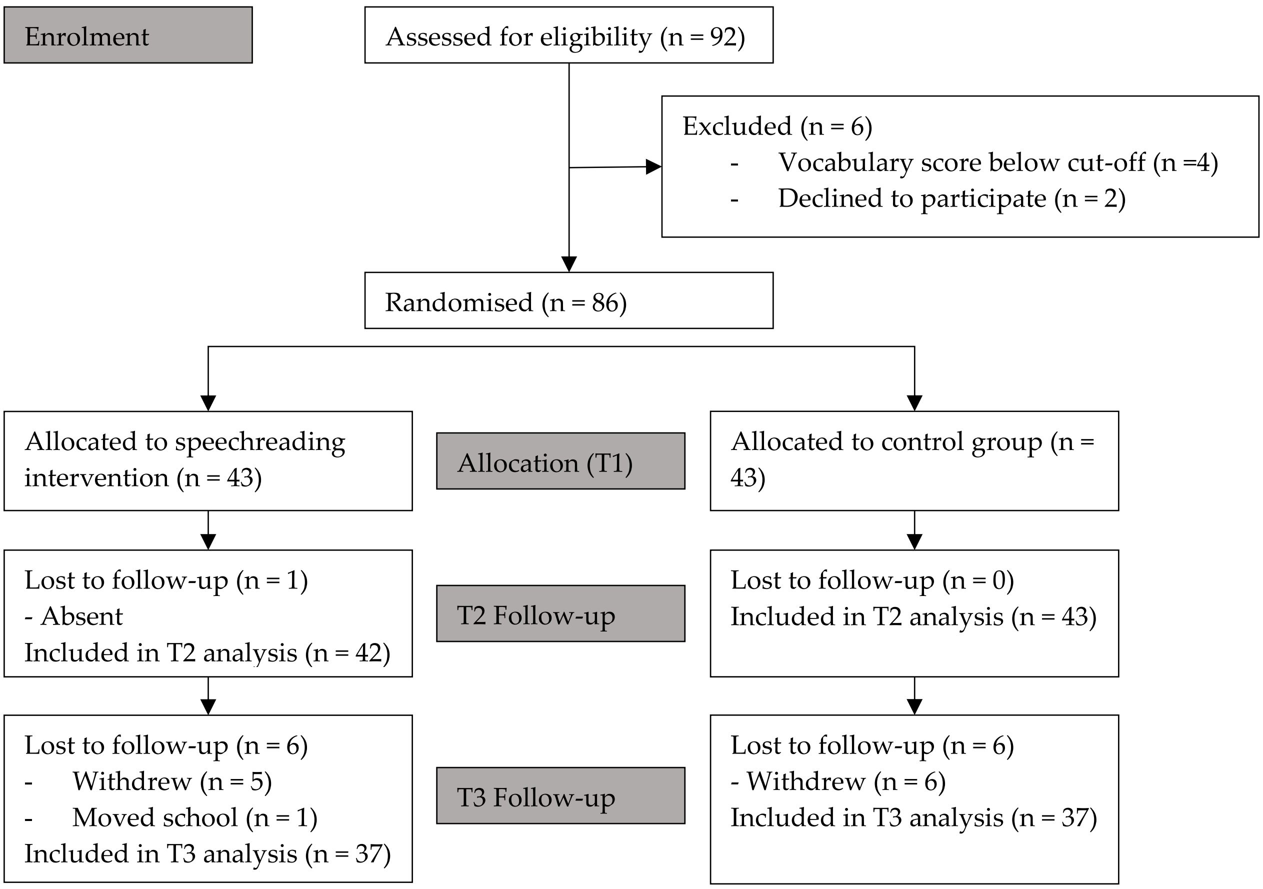

Figure 8. Flowchart documenting the movement of participants through the different stages of the trial.

8.1.3 Assessments

All children were assessed at baseline (T1), immediately after the intervention time (T2) and 3 months after the intervention (T3) on vocabulary, speechreading, phonological awareness and reading measures. All these measures included 20 words from the intervention (trained words) and the matched list of untrained words. Having 20 words in each list provided a focussed set of training words whilst maintaining a sufficient number of training words to avoid too much repetition. The trained and untrained items were alternated trial by trial throughout the assessment tasks.

8.1.3.1 Stimuli

The two lists of 20 words were compiled from a total of 103 words from the STAR_D games (Pimperton et al., submitted; see Appendix A; word lists presented in Appendix B). Words that were consistently named incorrectly in the STAR_D trial were removed completely. The lists were matched on: number of phonemes, letters and syllables; frequency (KF: Kucera & Francis, 1967 – count of words from a database of just over million words); the visibility of the words (proportion correct on a speechreading task – data from young deaf children; Pimperton et al., submitted) and the name agreement of the pictures (data from young hearing children), as shown in Table 6. The visibility and name agreement data were collected for the STAR_D project. One list was then selected as the training set based on the frequency of presentation across different levels of the STAR_D game (trained words). The images to represent each word were drawn by Simon Basher (http://www.basherworld.com/).

Table 6. Characteristics of the trained and untrained words used the study

	Untrained items		Trained items	
	M (*SD*)	**Range**	**M (*SD*)**	**Range**
Phonemes[a]	3.95 (*1.32*)	2-7	3.75 (1.33)	2-7
Letters[a]	5.00 (*1.41*)	3-9	4.95 (1.50)	3-9
Syllables[a]	1.40 (*0.68*)	1-3	1.25 (0.55)	1-3
Frequency (KF)[b]	73.72 (*149.55*)	1-591	68.39 (*102.77*)	4-431
Visibility[c]	0.27 (*0.21*)	0.04-0.79	0.25 (*0.16*)	0.05-0.51
Name agreement[c]	0.96 (*0.07*)	0.80-1	0.94 (*0.08*)	0.74-1

[a]Count of that sublexical component [b] Kucera & Francis (1967) count based on just over 1 million words [c]Proportion correct responses from pilot children

8.1.3.2 Vocabulary

The vocabulary task consisted of images of all 40 items from the trained and untrained set. The child was asked to name each item individually. If a child provided a similar label (e.g. 'bunny' instead of 'rabbit') they were prompted to provide a different label ("Can you think of another word for that?"). If the child couldn't name an item, the correct label was provided. The vocabulary task was only used at pre-test as a screening measure to ensure the children had a suitable proficiency in English.

8.1.3.3 Speechreading

Speechreading was measured with a single word task, including the trained and untrained items, and the Test of Child Speechreading Everyday Questions extension task.

8.1.3.3.1 Single word speechreading

The *primary outcome measure* was performance on the single word speechreading task. This speechreading task was made up of silent videos of models speaking all 40 items. The videos were ordered in terms of visibility, with the easiest words to speechread presented first and the trials alternated between trained and untrained items. The videos included the four models from the training game and the order of models was randomised. There were three practice trials at the start and after each of these the child was given verbal and visual feedback, with the correct answer circled on the screen. The first 20 trials were multiple-choice. After each video was played four images were presented at the four corners of the screen, including the target item and three unrelated images. Each child was asked to point to the image they thought matched the word in the video. The second half of the task was free-response. Each child was asked to guess what they thought the person had said in the video. They scored a point for each item correctly identified throughout the task, making a total of 40 points. If a child got 5 items in a row incorrect the task was stopped.

8.1.3.3.2 Sentence speechreading

Sentence speechreading was assessed with the 'Everyday Questions' subtest of the Test of Child Speechreading (Kyle et al., 2013, https://dcalportal.org/). Children could score a total of 62 points for the number of words they could correctly identify from the speechreading videos.

8.1.3.4 Phonological awareness

Phonological awareness was assessed at three levels: syllable blending, phoneme blending and phoneme deletion. The 40 trained and untrained items were divided across these three tasks as described below.

8.1.3.4.1 Blending

The blending tasks were modelled on the Clinical Evaluation of Language Fundamentals 4 (CELF 4; Semel, Wiig, Secord, & Langdon, 2006) tasks, using the same instructions and practice items. At the beginning of the tasks the experimenter said 'I will say a word very slowly. I want you to tell me what I'm saying' before breaking the word into either syllables or phonemes. The syllable-blending task included six items (3 trained) and the phoneme blending task included 14 items (7 trained). The time gap between the spoken phonemes was not measured, but experimenters were trained to break down the words and present them with a gap between each phoneme similar in length to the phonemes themselves.

8.1.3.4.2 Phoneme deletion

The sound deletion task was modelled on the YARC sound deletion task, using the same instructions and practice items. It included deletion of syllables, initial phonemes, final phonemes and phonemes from consonant clusters. There were 20 items (10 trained) plus 7 practice items. Feedback was only given on the practice items and one point was awarded for each test item. The phoneme deletion and vocabulary tasks were combined so that the child was shown an image, asked to name it and then asked to repeat the name without a specific sound. If the child got the vocabulary item wrong, they were provided with the correct answer before being asked to manipulate the sound.

8.1.3.5 Reading

In the reading task, each child was presented with the 40 words (20 trained and 20 untrained), shown on the screen four at a time. The words were ordered based on orthographic complexity and grapheme-phoneme correspondence and the trials alternated between trained and untrained items. Each child was asked to work through the screens reading the words aloud. No feedback was given. If 5 consecutive words were read incorrectly the task was stopped.

8.1.4 Intervention – adaptation

The training game was adapted from the STAR_D project speechreading and reading games to address the current question regarding the contribution of visual speech information to phonological awareness skills in young hearing children. Feedback from teachers and performance from the STAR_D project were used to adapt the training games to be suitable for 4-to-5-year-old hearing children. Teachers indicated that the deaf children in the STAR_D project enjoyed the space context of the games and the points where they were able to make choices about the games (e.g. choosing a captain character to play with) and commented on 10 minutes being an appropriate length of time for daily training. Therefore, both of these elements were maintained. The teachers had also indicated that the deaf children found trials that had phrases in too hard. These were therefore not included in the current STAR_H training.

The children were trained with the speechreading training game for 10 minutes a day, 5 days a week for 3 weeks. This set-up was derived from Melby-Lervåg and Hulme's (2010) study, which showed that short-term phonics training of 7 minutes a day over two weeks on blending and other phonological awareness tasks led to improvement on phoneme deletion, rhyme generation and serial recall of 10 trained words. Given that a moderate effect was seen in this study over two weeks of training, three weeks for the current study was considered an appropriate amount of time to see improvements on phonological awareness tasks for a small set of trained words.

Each session followed the structure outlined in Figure 9. The aim of the current study was to assess the contribution of visual speech information in the development of phonological awareness and thus it was considered appropriate to prioritise the phonological awareness aspect of the game. Therefore, each 10-minute session consisted of two speechreading games and two blending games, in order to explicitly teach blending through the visual modality. Blending was considered the most appropriate phonological awareness task to train as silent videos of broken words could be shown and then matched to pictures.

The set-up allowed an individual child to play a maximum of two sessions a day and 8 sessions a week in order to allow missed sessions to be caught up on subsequent days. In addition to the 20 training words (included in the pre- and post-tests) a list of 10 words were included in the training games to expose the children to a range of words. The additional set was selected based on phonological similarity to training words and was presented as videos and distractor items. A further 14 words were included at higher levels of the game in order to allow for closer matching between the targets and distractors (see below for description of game progression). Words from the untrained list were never presented as videos in the games but could appear as distractor pictures. The children played the games in a room with 1-4 other players at a time, each playing on their own touch-screen device (Microsoft-Surface Pro) via the internet. These sessions were supervised by the experimenter. In every game the trials only progressed once the child had selected the correct response. If a child responded incorrectly they had to re-watch the video before being able to select any of the response options.

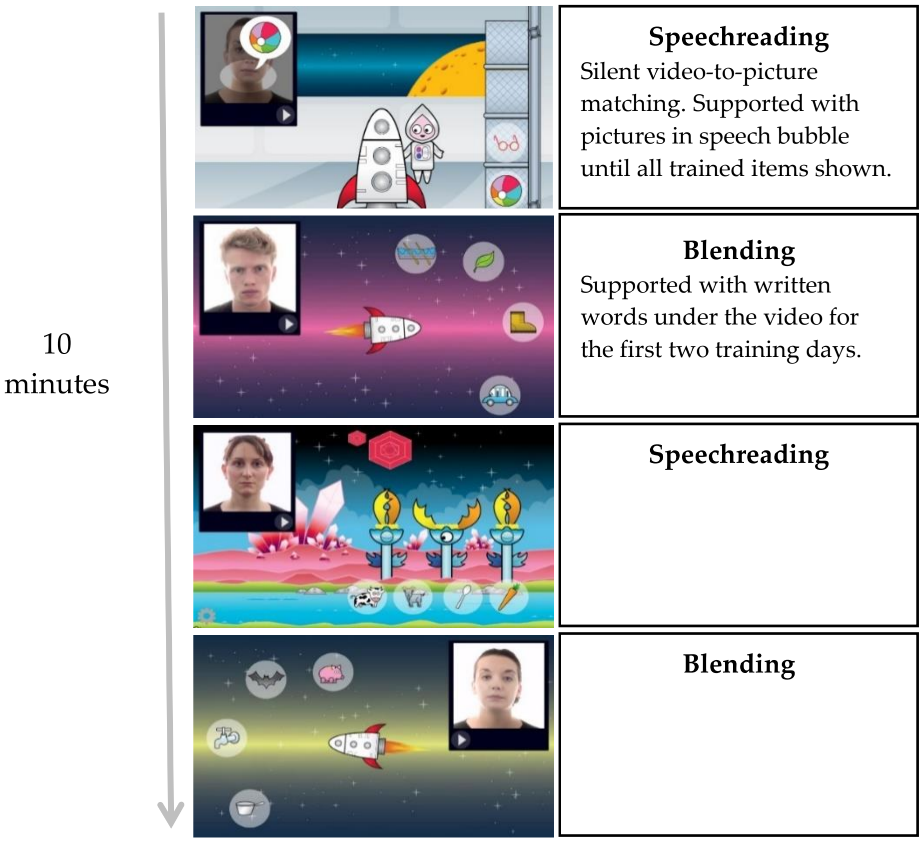

Figure 9. Schematic of each 10-minute training session. The first and third game of each day involved single word speechreading and selecting a matching image. The second and fourth games of each day involved blending silent videos of the model breaking down a word (e.g. ‘b-a-t’) and selecting the matching picture. The planet that the children worked on rotated through four different options in order to maintain interest.

The games progressed in two ways. First, the games progressed by changing the level of support the children received. On the first game of each day ("Pack the rocket") a video of a model saying a single word was presented, followed by a speech bubble showing what they said. The child then selected what the model had said from two pictures. Each speechreading trial was immediately followed by a paired trial where the picture from the previous trial was presented and the child watched two videos of a model speaking and selected the word that matched the picture. Once the child had seen all the training items in this introductory format the speech bubble was removed. Blending games were included in this training as the aim was to examine the contribution of visual speech information to phonological awareness skills. Therefore, it was considered appropriate to train a phonological awareness skill, such as blending, within the speechreading training. For the blending ("Space junk") games the children saw a model 'sound out' a word (e.g. the model said 'b-a-t') and the child had to select the matching image (e.g. of a bat). The children were supported on the blending games for the first two days of training with the word written underneath the video of the model and each letter highlighted as it was pronounced. The writing was removed on day three and only shown on the first trial of each blending game to remind the child of the task.

The second form of progression was in the type of distractors presented. The games used the algorithm from the STAR games, which allowed the difficulty level to adapt according to the child's performance. The distractors presented were initially visually distinct from the target item and progressively increased in similarity when the child achieved a threshold for success at that level.

The blending games progressed from having the whole word spoken after the broken-down word (e.g. 'b-a-t-bat') to just having the broken-down word (e.g. 'b-a-t') when the child had been successful in identifying that word at the first level. The blending games included only the 20 trained items that were included in the pre- and post-tests. If a child completed both levels of the blending games with this set of items, the additional items were included into the blending games so they could continue playing. The same algorithm was carried across both blending games that were played each day so that progress in one game led to increased difficulty in both games.

8.1.5 Control group

The control children were only seen by the experimenter at pre- and post-tests. They did not receive any intervention but received half an hour of phonics training in school every day as is standard. Children in Reception receive daily phonics lessons, which include recognition of individual letters and digraphs and blending of words. The classes are aimed at preparing the children to be able to decode words by the end of year 1 (aged 6), which is assessed by the phonics screening check involving reading 20 words and 20 pseudowords (https://www.gov.uk/education/phonics).

8.1.6 Statistical methods

Differences in performance between the speechreading intervention group and the control group at T2 and T3 were assessed using ANCOVAs. The dependent variable in each model was the performance on an assessment at either T2 or T3, the covariate was the performance on that assessment at T1 and the fixed factor was the group (intervention or control). Performance on each of the measures was considered to be normally distributed based on visual inspection of Q-Q plots. The assumption of equality of slopes was assessed by including an interaction term in the ANCOVA models between the covariate and the group.

Within the intervention group, differences in performance on trained and untrained items was assessed using a series of paired t-tests, comparing performance between trained and untrained items at each timepoint, and assessing improvement for each item type. The t-tests were Bonferroni corrected for multiple comparisons (α = .0125). When a difference in performance was observed post intervention, the interaction between time and item type was assessed further t-tests to determine whether a greater improvement was made on trained than untrained items.

8.2 Results

Assessment data were collected from 86 children at T1 (43 speechreading intervention, 43 control), 85 children at T2 (42 speechreading intervention, 43 control), and 74 children at T3 (37 speechreading intervention, 37 control) (see Figure 8 – CONSORT diagram).

The mean time between T1 and T2 assessments was 4.02 weeks (*SD* = 0.29, range: 3-5 weeks). The time between T1 and T2 assessments did not differ between the speechreading intervention group (*M* = 3.99, *SD* = 0.29, range: 3-5 weeks) and the control group (*M* = 4.05, *SD* = 0.29, range: 3-5 weeks), $t(83)$ = 1.00, p = .319, [95% CI –0.06, – 0.19]. The mean time between T1 and T3 assessments was 17.36 weeks (*SD* = 1.02, range: 16-21 weeks). The time between T1 and T3 assessments did not differ between the speechreading intervention group (*M* = 17.37, *SD* = 1.14) and the BAU control group (*M* =

17.34, *SD* = 0.92), *t*(73) = –0.15, *p* = .880, [95% CI –0.51, – 0.44]. Adherence to the intervention was high. Forty-two of the 43 children in the intervention group completed all 15 training sessions. One child completed 14 of the 15 training sessions.

Table 7 shows the means, standard deviations and Cohen's *d* for each group at baseline (T1), immediate post-test (T2) and three-month follow-up (T3). Cohen's *d* was calculated as the difference in gains between groups from T1 to each post-test divided by the pooled standard deviation at T1.

Table 7. Descriptive statistics for the intervention and control groups

	Intervention		BAU Control		
	M	**SD**	**M**	**SD**	**Cohen's *d***
Vocabulary (0-40)					
T1	38.33	1.91	37.95	2.48	
Single word speechreading (0-40)					
T1	4.83	3.59	5.02	4.34	
T2	10.05	1.40	6.30	1.78	.99
T3	7.92	5.96	6.11	4.51	.50
TOCS extension words score (0-62)					
T1	4.38	5.49	5.33	6.13	
T2	7.38	7.65	7.53	6.99	.14
Syllable blending (0-6)					
T1	4.43	2.20	4.98	1.79	
T2	5.55	1.23	5.23	1.73	.43
Phoneme blending (0-14)					
T1	10.67	3.53	10.49	4.01	
T2	11.74	2.86	11.37	4.02	.05
Phoneme deletion (0-20)					
T1	6.50	4.59	6.30	4.95	
T2	8.29	4.80	7.72	5.44	.08
T3	10.38	4.83	10.11	4.93	.01
Single word reading (0-40)					
T1	8.81	9.95	7.16	7.79	
T2	11.79	11.84	10.65	9.63	−.06
T3	20.05	11.81	17.53	12.36	.10

Note. At T1 n = 86, at T2 n = 85, at T3 n = 74.

8.2.1 Planned analyses

8.2.1.1 Single word speechreading

An ANCOVA of T2 single word speechreading scores, controlling for baseline single word speechreading scores, showed an advantage for the intervention group (difference between groups = 3.88 [95% CI 1.74, 6.03]; t = 3.60, $p < .001$; standardised mean difference between groups = .99).

Within the speechreading intervention group, paired t-tests revealed a significant improvement on single word speechreading on both trained and untrained words (trained: $t(41) = 5.56$, $p < .001$, $d = 0.86$; untrained: $t(41) = 5.20$, $p < .001$, $d = 0.80$). The trained children performed significantly better on the trained items than on the untrained items at T2 ($t(41) = 2.76$, $p = .009$, $d = 0.43$) and there was no difference in performance at pre-test ($t(41) = –0.40$, $p = .691$, $d = –0.06$). A paired t-test on the difference scores from T1 to T2 revealed a significantly greater improvement on the trained words than the untrained words ($t(41) = 2.67$, $p = .007$, $d = 0.41$).

8.2.1.2 Sentence speechreading

An ANCOVA of T2 TOCS extension scores, controlling for baseline TOCS extension scores, showed no significant difference between the speechreading intervention and BAU control groups (difference between groups = 0.51 [95% CI -2.14, 3.16]; $t = 0.381$, $p = .704$; standardised mean difference between groups = .14).

8.2.1.3 Phonological awareness

The pre-registration specified that performance on the three phonological awareness tasks would be combined if they showed a strong correlation ($r > .60$) with each other. Syllable blending was excluded as a measure due to ceiling effects (see Table 7). Phoneme blending and phoneme deletion scores did not correlate with each other sufficiently (T1: $r = .53$; T2: $r = .54$) and so were not combined. The planned analyses (ANCOVA and t-tests) are therefore reported for each measure independently below.

8.2.1.3.1 Phoneme blending

An ANCOVA of T2 phoneme blending scores, controlling for baseline phoneme blending score, showed no significant difference between the speechreading intervention and BAU control groups (difference between groups = 0.21 [95% CI –0.34, 0.77]; $t = 0.76$, $p = .449$; standardised mean difference between groups = .05).

The assumption of equal slopes was not met for the phoneme blending task, the interaction term between the group and covariate was significant (unstandardised slope = –0.20 [95% CI –.35, –.06]). This corresponds to a shallower slope for the intervention group, meaning that the intervention led to a greater improvement in phoneme blending for children who initially had lower scores on this measure. Follow-up tests indicated that the groups did not differ at post-test at the mean of the covariate (difference between groups = 0.22 [95% CI –.32, .75]; $t = 0.80$, $p = .426$; standardised mean difference between groups = .06). However, for children scoring 1 standard deviation below the mean at pre-test (the covariate) there was a significant advantage for the intervention group (difference between groups = 0.97 [95% CI .21, 1.74]; $t = 2.53$, $p = .013$; standardised mean difference between groups = .26) at post-test. These results must be interpreted with caution as there were few children scoring at the lower end on this measure, but this pattern suggests that the speechreading training was effective for children with low initial phoneme blending scores.

Within the speechreading intervention group, paired t-tests revealed a significant improvement on phoneme blending on both trained and untrained words (trained: $t(41) = 3.74$, $p < .001$, $d = 0.58$; untrained: $t(41) =$

3.04, $p = .002$, $d = 0.47$). The trained children performed significantly better on the trained items than the untrained items at T2 ($t(41) = 3.57$, $p = .001$, $d = 0.55$). However, there was also a significant difference in performance at pre-test between trained and untrained words ($t(41) = 3.02$, $p = .004$, $d = 0.47$).

8.2.1.3.2 <u>Phoneme deletion</u>

An ANCOVA of T2 phoneme deletion scores, controlling for baseline phoneme deletion score, showed no significant difference between the speechreading intervention and BAU control groups (difference between groups = 0.39 [95% CI –0.89, 1.68]; $t = 0.605$, $p = .547$; standardised mean difference between groups = .08).

Within the speechreading intervention group, paired t-tests revealed a significant improvement on phoneme deletion on both trained and untrained words (trained: $t(41) = 2.65$, $p = .006$, $d = 0.41$; untrained: $t(41) = 3.85$, $p < .001$, $d = 0.59$). The trained children performed significantly better on the trained items than on the untrained items at T2 ($t(41) = 2.79$, $p = .008$, $d = 0.43$). However, there was also a significant difference in performance at pre-test between trained and untrained words ($t(41) = 3.87$, $p < .001$, $d = 0.60$).

8.2.1.4 Reading

An ANCOVA of T2 single word reading scores, controlling for baseline single word reading score, showed no significant difference between the speechreading intervention and BAU control groups (difference between groups = –0.69 [95% CI -2.54, 1.15]; $t = –0.75$, $p = .456$; standardised mean difference between groups = –.06).

Within the speechreading intervention group, paired t-tests revealed a significant improvement on single word speechreading on both trained and untrained words (trained: $t(41) = 4.07$, $p < .001$, $d = 0.63$; untrained: $t(41) = 3.90$, $p < .001$, $d = 0.60$). There was no difference in performance on trained and untrained items at T2 ($t(41) = –0.21$, $p = .838$, $d = –0.03$). However, there was a significant difference in performance at pre-test between trained and untrained words ($t(41) = -2.26$, $p = .029$, $d = –0.35$).

8.2.2 Exploratory analyses – three-month follow-up

Given the improvement on speechreading scores for the intervention group over the control group at T2, the children were followed up three months post intervention (T3) to determine whether the improvement on speechreading skill was maintained and whether there was a knock-on effect on phonological awareness and reading ability. The analyses reported at T2 were repeated with T1 and T3 scores.

8.2.2.1 Speechreading

An ANCOVA of T3 single word speechreading scores, controlling for baseline single word speechreading scores, showed a significant main effect of group (difference between groups = 2.46 [95% CI 0.42, 4.50]; $t = 2.40$, $p = .019$; standardised mean difference between groups = .50).

Within the speechreading intervention group, paired t-tests revealed a significant improvement from baseline to T3 on single word speechreading on both trained and untrained words (trained: $t(37) = 3.63$, $p < .001$, $d = 0.59$; untrained: $t(37) = 3.76$, $p < .001$, $d = 0.61$). There was no difference in performance at T3 on trained and untrained items ($t(37) = -1.62$, $p = .114$, d = 0.26).

8.2.2.2 *Phoneme deletion*

An ANCOVA of T3 phoneme deletion scores, controlling for baseline phoneme deletion score, showed no significant difference between the speechreading intervention and control groups (difference between groups = 0.07 [95% CI –1.38, 1.52]; $t = 0.10$, $p = .923$; standardised mean difference between groups = .01).

Within the speechreading intervention group, paired t-tests revealed a significant improvement from baseline to T3 on phoneme deletion on both trained and untrained words (trained: $t(37) = 4.51$, $p < .001$, $d = 0.73$; untrained: $t(37) = 6.59$, $p < .001$, $d = 1.07$). There was no difference in performance at T3 on trained and untrained items ($t(37) = 0.63$, $p = .534$, d = 0.10).

8.2.2.3 *Reading*

An ANCOVA of T3 single word reading scores, controlling for baseline single word reading score, showed no significant difference between the speechreading intervention and control groups (difference between groups = 0.14 [95% CI –3.25, 3.53]; $t = 0.08$, $p = .934$; standardised mean difference between groups = .10).

Within the speechreading intervention group, paired t-tests revealed a significant improvement from baseline to T3 on single word speechreading on both trained and untrained words (trained: $t(37) = 9.26$, $p < .001$, $d = 1.50$; untrained: $t(37) = 9.32$, $p < .001$, $d = 1.51$). There was no difference in performance at T3 on trained and untrained items ($t(37) = –0.17$, $p = .862$, $d = 0.03$).

8.3 Discussion

The current randomised experiment adapted a speechreading training intervention to investigate whether speechreading can be trained over 3 weeks in young hearing children and whether this training boosts phonological awareness over and above the day-to-day phonics training they receive at school.

8.3.1 Speechreading

The first hypothesis that speechreading training would lead to improvements in speechreading was in general supported. The speechreading intervention group performed better than the business-as-usual control group on the single word speechreading post-test when controlling for baseline scores on the same test. In addition, within the speechreading intervention group, the children improved on both trained and untrained words, but showed a greater improvement on trained words. These effects were maintained at the 3-month follow-up.

The results challenge the idea that speechreading is a fixed skill that cannot be trained (Montgomery & Sylvester, 1984; Summerfield, 1992). There is evidence that speechreading can be trained in deaf adults (Bothe, 2007; Lonka, 1995; Walden et al., 1977), hearing adults (Bernstein et al., 2001;

Blumsack et al., 2007). The Speechreading Training and Reading (STAR) programme has been shown to be effective in improving speechreading in deaf children (Pimperton et al., submitted) and the current study shows that it can also be trained in hearing children. This suggests that improvements in speechreading can arise as a result of short-term training regardless of the extent to which the individual relies on visual speech information to access spoken language in daily life.

Although improvements were seen on the single word speechreading task, the speechreading training did not result in improved performance on the Test of Child Speechreading (TOCS) Everyday Questions extension task. In this task the children were asked to repeat a full speechread everyday question, such as 'How old are you?'. Unlike the single word speechreading task, the TOCS extension had different talkers to the training games and has a free-response format rather than multiple choice. The differences in task type may explain the finding that improvements on the single word speechreading task as a result of the intervention were not also seen on the TOCS Everyday Questions extension task. First, previous research has shown that hearing adults are better at speechreading familiar faces than unfamiliar faces (Lander & Davies, 2008) and therefore it is possible that the training would not extend to untrained talkers. Second, the multiple-choice format of the single word speechreading task meant that the children's options for response were narrowed down. Therefore, in contrast to the TOCS extension task, they did not necessarily have to identify the whole word in order to select the correct picture, just some of the phonemes. It may be that even if the children made improvements in identifying phonemes this was not sufficient to observe improvements in the free-response task. In addition, improvements on the single word speechreading may be partly explained by familiarity with the format as it matched the format of the training games. This is one limitation of using a BAU control group rather than an active control group, for whom familiarity with the testing format is similar to that of the intervention group. Unfortunately, due to time and resource constraints, an active control group was not possible for this proof-of-principle study.

Sentence-level speechreading ability is generally worse than single word speechreading ability in deaf and hearing adults and children (Kyle et al., 2013; Mohammed et al., 2006). In the TOCS extension task the children were reminded regularly that they were looking for a question. However, the children tested here were very young (4-5 years old). Many often provided a statement response or a single word indicating that they may not have understood what a question was. Therefore, the improvements seen on single word speechreading but not on sentence-level speechreading may be due to sentences being more linguistically complex stimuli.

The improvements seen on single word speechreading but not on sentence-level speechreading as a result of the intervention in hearing children are in contrast to the assessment of the STAR programme with deaf children (STAR_D, Pimperton et al., submitted). The deaf children did not show improvements on the TOCS single word speechreading test as a result of the intervention, which had untrained talkers and untrained stimuli. However, they did show improvement on the STAR_D in-game assessments with trained talkers and trained items at the immediate post-test, as did the hearing children in the current study. Unlike the hearing children, the deaf children did not show improvement on the untrained words but did improve on the TOCS Everyday Questions extension task, which had both untrained talkers and sentence-level stimuli.

One reason for the difference in results between the current study and the STAR_D project may be that the training was over 3 times longer in the STAR_D project (12 weeks, 48 sessions) than in the current study (3 weeks, 15 sessions). In addition, the children in the STAR_D training played some games that used sentences, for example "Find the sheep.". It may be that the longer period of training and the exposure to sentences in the training is necessary to lead to improvements in the more complex sentence-level speechreading as tested in the TOCS Everyday Questions extension task.

8.3.2 Phonological awareness

The second hypothesis that speechreading training would lead to improvements in phonological awareness skill was not well supported. There were no differences in performance between the speechreading training group and the control group on post-tests of either phoneme blending or phoneme deletion when controlling for baseline scores. Follow-up analyses 3 months later also showed no improvement in phoneme deletion scores as a result of the intervention.

Within the speechreading intervention group the children made improvements on the phonological awareness tasks for both trained and untrained items. However, phonics instruction is a dominant aspect of the curriculum for this age group as recommended in the Rose report (Rose, 2006) and so both the speechreading intervention and the BAU control groups made huge improvements from baseline to both post-test timepoints.

Any improvements on the phonological awareness tasks as a result of the speechreading intervention are likely to be small and therefore may not have been detected in the context of the huge gains made. In addition, pilot work revealed that at the beginning (September) of Reception the children could not attempt the phonological awareness tasks but when tested for this project at the beginning of their second term in school (January-March) many were already at ceiling on the phoneme blending task. This made it harder to observe any improvement on the phoneme blending task.

Although many children make rapid progress, many children struggle with phonological awareness skills. It is likely to be specifically these children that may benefit from their attention being brought to visual speech in order to augment and support their understanding of the sublexical structure of words through sound alone (Heikkilä et al., 2016; Knowland et al., 2016; Mohammed et al., 2006). The contribution of speechreading to phonological awareness skills may therefore be dependent on the child's initial phonological awareness ability. The children in the intervention group who started off with low scores on the phoneme blending task performed better at post-test than the BAU control group children with low baseline scores. This suggests, in line with our predictions, that visual speech information is contributing to phonological representations (Kyle & Harris, 2011; Mohammed et al., 2006), and can provide an additional route to developing phonological skills for those with poorer initial performance. The children in the speechreading training group played two silent phoneme blending games per training day, so this interaction may indicate that practicing blending in one modality (visual) may boost performance in a second modality (auditory). However, it is also possible that small differences in the very few poorest performing children (10/85 children scored more than 6.8, one standard deviation below the mean of the group) were exaggerated in the analysis by the clustering of children performing at ceiling on the phoneme blending task (55/85 children scored more than 10/14). Future studies targeting those children with poorer phoneme blending skills will allow these two possible interpretations to be distinguished. Such studies focussing on this group will face challenges of recruitment, as it would require screening of whole classes in order to identify children who are struggling with phonics at this young age.

An effect on phonological awareness may not have been observed as the effect on the single word speechreading task was moderate, and therefore may not have been large enough to lead to improvements in phonological awareness. In addition, despite the training showing generalised effects to untrained words within the same task format, the training did not generalise to sentence speechreading with unfamiliar models. Therefore, the improvement in speechreading may not have been sufficient for the children to apply this to make use of visual speech information during daily audiovisual speech perception to support the development of phonological representations and skills.

To understand more specific effects of training on phonological awareness skills it was hypothesised that the children in the speechreading training group would be better at phonological awareness tasks with the trained items than the untrained items at post-test. Although this was found, there was also a significant difference in performance on the trained and untrained items at baseline, making the results difficult to interpret. This may be because although the trained and untrained words were matched on several measures (e.g. number of phonemes, letters and syllables; frequency; the visibility of the words; and the name agreement of the pictures) they were matched on the mean of the different measures rather than item by item. In the phoneme blending task the imbalance in performance at pre-test may have been a result of the final few items where there were three items in the untrained list that required blending of consecutive consonants but only one item in the trained list that required this. Many children found blending consonant clusters harder than consonant-vowel-consonant blending (e.g. s-t-ar vs. s-o-ck). In the phoneme deletion task, the difference in baseline performance on the trained and untrained items may similarly be explained by a few difficult items. A couple of untrained items had 3 syllables and generally children performed worse on these items than on the neighbouring trained items. The difference in performance at pre-test on the phonological awareness tasks limited the ability to detect specific improvements in phonological awareness skills on trained words as a result of the intervention. However, regardless of the difference in performance on trained and untrained items at pre-test, the improvement made was similar across both word types. Therefore, there does not seem to be a training-specific improvement on phonological awareness tasks.

In the STAR_D project the deaf children also did not show improvements on the phonological awareness task as a result of the intervention but they did improve in their speech output. Speech output was used as a measure of the quality of their underlying phonological representations. In hearing children around 4-5 years old, the quality of speech production is often still being refined, even for children with full access to auditory speech to model their own speech on and for feedback on their own speech production. Therefore, the quality of speech output may also be an informative measure to use in hearing children to assess the effects of the speechreading intervention on underlying phonological representations (Carroll et al., 2003). Video data were collected in the current study to allow for future analysis of speech output.

8.3.3 Reading

The final hypothesis was that if speechreading training resulted in improvements on phonological awareness tasks there would also be related improvements on reading. However, this hypothesis was not supported. The speechreading training group did not differ in performance on the reading post-tests from the BAU control group at the immediate follow-up or the 3-month follow-up. Similarly, within the speechreading training group there was no difference in performance on trained and untrained words at either post-test, although there was a difference at pre-test. The improvement observed from baseline to post-test reading scores on both trained and untrained items is likely to be general improvement unrelated to the intervention. It is not surprising that there were no differences in reading as a result of the intervention as single word reading is strongly predicted by phonological awareness ability in hearing children, and this did not improve as a result of the intervention (Caravolas et al., 2012; Castles & Coltheart, 2004; Hatcher et al., 2004; Hulme et al., 2002). As with the phonological awareness tasks, the children in Reception are making huge gains in their reading skills at this stage and so any small improvement that may have resulted from the intervention may have been lost in this context.

The current results are in contrast to previous studies that have found speechreading predicts later single word reading ability in both deaf and hearing children (Kyle, 2015; Kyle et al., 2016; Kyle & Harris, 2010). In addition, they lend only weak support to the structural equation models presented in Chapters 6 and 7 that suggest that the relationship between speechreading and reading is mediated by phonological awareness skills in both deaf and hearing children in a concurrent dataset. In Chapters 6 and 7 it was hypothesised that one explanation for these models is that phonological representations are multimodal, representing what a person hears and sees when someone is talking as well as their own speech production. This explanation suggests that speechreading is causally related to phonological awareness by building more robust phonological representations (Kyle & Harris, 2011; Mohammed et al., 2006). Further explanations for the discrepancy in results between the structural equation model in Chapter 7 and the current intervention study will be discussed in detail in Chapter 9.

This study has demonstrated the efficacy of the STAR intervention in young hearing children, showing that speechreading can be trained in hearing children as well as deaf children over a relatively short period of time. Reading is a fundamental skill for young children to develop in order to access the rest of education. Given the relationship between phonological skills and reading development in hearing children it is important to understand what information contributes to the development of phonological representations and awareness. Visual speech perception is suggested to contribute to phonetic discrimination in hearing infants (Teinonen et al., 2008), which in turn has been shown to relate to language development in hearing children (Kuhl et al., 2005). Therefore, understanding the role of visual speech information in the development of phonological skills is not only relevant to education for deaf children but also for hearing children.

9 General discussion

The aims of this thesis were to investigate how deaf and hearing children access visual speech information and to investigate the nature of the relationship between speechreading, phonological awareness and reading in both deaf and hearing children. This chapter will consider previous evidence and the findings from Chapters 5-8 to address the following questions:

1. *How do eye movements during speechreading relate to speechreading skill in deaf and hearing children?*

2. *Is there a relationship between speechreading, phonological awareness and reading in deaf and hearing children?*

3. *What is the causal direction of any relationships between speechreading, phonological awareness and reading?*

9.1 How do eye movements during speechreading relate to speechreading skill in deaf and hearing children?

Children born deaf must rely to a greater extent on visual input to access spoken language than hearing children. Deaf adults show an advantage on speechreading measures compared to hearing adults (Bernstein et al., 2000; Mohammed et al., 2006; Pimperton et al., 2017). However, previous studies have shown that deaf and hearing children show very similar speechreading ability (Kyle et al., 2013; Kyle & Harris, 2006). Chapter 5 supported these previous studies showing that, despite greater reliance on visual information for understanding speech, 5-to-8-year-old deaf children showed similar levels of speechreading ability to their hearing peers.

Given the similarity in speechreading skill in deaf and hearing children and the relationship between speechreading and reading ability in both groups it is of interest to consider how both deaf and hearing children access silent speech information. Chapter 5 used eyetracking during a sentence speechreading task in deaf and hearing children. The deaf and hearing children looked at the mouth for a similar amount of time. To our knowledge this is the first time this issue has been addressed in children whether deaf or hearing. Our findings are in line with previous research showing that hearing adults look at the mouth when auditory information is compromised during audiovisual speech perception in noise as well as when speechreading (Lansing & McConkie, 2003; Vatikiotis-Bateson et al., 1998).

For both groups of children, the time they spent looking at the mouth of the speaker in the silent videos correlated with their speechreading ability. This is in contrast to two previous studies with hearing adults that did not find a relationship between mouth-focus and speechreading (Lansing & McConkie, 2003; Wilson et al., 2016). The contrast in results between the current and previous studies may be due to the age of the participants. It is possible that differences between deaf and hearing children in their gaze behaviour, and how this relates to speechreading ability, emerge later in development than for the ages studied here. In addition to the predicted relationship between mouth-focus and speechreading performance, many children started each trial looking at the eyes before the speech started, shifted their gaze to the mouth during speech and returned to look at the eyes once the speech had finished. Both groups showed this social-tuning pattern, which was previously observed in hearing adults during a speechreading task (Lansing & McConkie, 2003). Although deaf children did not employ the social-tuning pattern more than hearing children, this pattern was more strongly related to speechreading skills in deaf than hearing children. It was argued in Chapter 5 that this gaze pattern reflects more general underlying language skills, which may explain why it also relates to deaf children's reading ability.

These results suggest that deaf and hearing children are very similar in their gaze behaviour when watching silent speech and in how this relates to their speechreading ability. The similar eye movement patterns between the deaf and hearing children in the speechreading task suggest that these two groups of children may be able to use visual speech information in a similar way to contribute to spoken word representations. In addition, it suggests that the mouth is an important source of speech information as those who showed greater attention to the mouth were able to repeat more words on the speechreading task.

9.2 Is there a relationship between speechreading, phonological awareness and reading in deaf and hearing children?

To address this question we will consider the relationship between speechreading and reading; whether phonological awareness relates to reading ability in deaf children; and whether the concurrent relationship between speechreading and reading is mediated by phonological awareness in deaf and hearing children.

9.2.1 A relationship between speechreading and reading in deaf and hearing children

Speechreading and reading ability have been repeatedly found to relate to each other concurrently in both deaf and hearing children (Arnold & Köpsel, 1996; Harris et al., 2017b; Kyle et al., 2016; Kyle & Harris, 2006) and longitudinally in deaf children (Harris et al., 2017b; Kyle & Harris, 2010, 2011). In Chapter 6 I reported a structural equation model using data from deaf children. These data were the baseline data from the STAR_D project (see Appendix A). The results replicated previous studies showing that speechreading, measured using the Test of Child Speechreading Everyday Questions extension task (Kyle et al., 2013), relates to concurrent single word reading ability in young deaf children (r = .58 and .63 with early word and single word reading tests respectively). In Chapter 7 young hearing children (5-to-8-year-olds) completed the same sentence-level speechreading task as the deaf children and some of the same single word reading tasks. As with the deaf children and in line with previous studies (r = .31, Kyle et al., 2016; r = .58, Kyle & Harris, 2010), speechreading ability correlated with concurrent reading ability in the young hearing children (r = .49 and .56 with early word and single word reading tests respectively). A previous study with hearing adults suggested a relationship between speechreading and reading in dyslexic participants but not in non-dyslexic participants (Mohammed et al., 2006). One reason why speechreading may relate to reading ability in hearing children but not in hearing adults (Mohammed et al., 2006) is that there is less variability in the reading ability of hearing adults than in children's reading skills. With little variability in reading scores, it may be difficult to observe a correlation with speechreading ability.

9.2.2 Does phonological awareness relate to reading ability in young deaf children?

Phonological awareness is known to be an important predictor of reading ability in hearing children (see Melby-Lervåg, Lyster, & Hulme, 2012). However, the results from previous studies with young deaf children are less clear (Campbell & Wright, 1988; Conrad, 1979; Dyer et al., 2003; Hanson & Fowler, 1987; Harris & Beech, 1998; Kyle & Harris, 2006; Leybaert & Alegria, 2003; Mayberry et al., 2011). Chapter 6 showed that in a representative sample of young deaf children, phonological awareness scores (onset and rime judgement) positively correlated with single word reading skills (*r*'s between .50 and .60). As well as a relationship between the raw scores on the measures of phonological awareness and reading, the latent variables for each of these factors in the structural equation model with deaf children also showed a strong relationship with each other. Therefore the results in Chapter 6 suggest that the relationship between phonological awareness and reading ability in young deaf children in this study is robust to measurement error.

In the light of the given mixed findings from previous studies, there are some methodological factors that are worth noting in regard to the relationship between phonological awareness and reading in the deaf children observed in Chapter 6. First, the current data with deaf children represents a large sample compared to many previous studies. However, when working with the deaf population, a large sample inevitably brings variability. Twenty of the children used spoken English as their preferred communication in school, 33 used a mixture of speech and signing with varying degrees of proficiency with each, and 13 of the children used only British Sign Language (BSL) in school. As the majority of the children (46/66) in this study used BSL to some extent it suggests that the relationship between phonological awareness and reading in deaf children is not limited to those who only use a spoken language. Therefore, the relationship between phonological awareness and reading is robust enough to be observed despite variability in language use in this sample. However, care should be taken when applying these results to individual subsamples of deaf children (e.g. BSL only, spoken language only or mixed).

A second methodological consideration is that almost all of the deaf children studied in Chapter 6 (n = 60) had either bilateral cochlear implants or hearing aids. The proportion of children in our study with cochlear implants is in line with the proportion in the UK as a whole (Raine, 2013). Given the relatively recent introduction of universal newborn hearing screening in the UK (2006), and the constant development of technology, the children in Chapter 6 are likely to have earlier and greater access to the auditory component of spoken language than deaf children involved in previous published studies. Therefore, it is likely that they were able to use auditory information to a greater extent than children in previous studies to help develop their phonological representations and therefore support their single word reading.

Longitudinal studies in the field of deafness and reading are rare. Of the few that have been reported, they found that despite a concurrent relationship between reading and phonological awareness, phonological awareness skills did not predict later reading ability (Harris et al., 2017b; Kyle & Harris, 2010). Kyle & Harris (2010) reported that early phonological awareness skills did not predict later reading skills but earlier reading ability did predict later phonological awareness. Similarly, others have found that deaf children develop their syllabic awareness after learning to read (Harris & Moreno, 2004). Therefore, it is possible that learning to read gives deaf children insight into the sublexical structure of speech, allowing them to subsequently complete the manipulations required in phonological awareness tasks. The direction of this relationship and the development of phonological awareness in deaf children will be discussed further below.

9.2.3 Is the concurrent relationship between speechreading and reading mediated by phonological awareness in deaf and hearing children?

The structural equation model in Chapter 6 based on data from young deaf children showed that the direct relationship between speechreading and reading ability was fully mediated by phonological awareness scores. This supports the proposal that there is an indirect relationship between speechreading and reading in deaf children via phonological awareness (Kyle, 2015; Kyle & Harris, 2010). Given the role of visual speech perception in phoneme discrimination and language development in hearing infants and children (Erdener & Burnham, 2018; Jerger et al., 2018; Weatherhead & White, 2017; Weikum et al., 2007), Chapter 7 investigated a similar model in hearing children. As with the deaf children, the direct relationship between speechreading and reading skills in the hearing children was fully mediated by their phonological awareness scores. This is the first demonstration of this relationship in young hearing children. Different measures were used in the structural equation models with deaf and hearing children, and therefore the models were not directly comparable. However, the two models showed strikingly similar relationships between speechreading, phonological awareness and reading ability, suggesting that the relationships may be similar for both deaf and hearing children.

First, we consider the pattern observed in deaf children. Kyle (2015) has previously proposed that in deaf children speechreading ability relates to reading ability both via phonological awareness and via a route independent of phonological awareness. A plausible route independent of phonological awareness is that speechreading improves deaf children's vocabulary, which in turn is a positive predictor of reading ability. In the model in Chapter 6 with data from deaf children the direct pathway between speechreading and reading was reduced to a trivial size when phonological awareness was added into the model. This supports the mediated pathway via phonological awareness. However, this does not refute the suggestion that there is also a relationship between speechreading and reading ability independent of phonological awareness at some stage of reading development in deaf children. Kyle (2015) proposed that the independent relationship between speechreading and reading precedes the development of the relationship via phonological awareness. Kyle (2015) did not specify whether the relationship independent of phonological awareness remains after early reading and phonological awareness skills have been established or whether the route via phonological awareness replaces it. In the model in Chapter 6 the mean age of deaf children was 6 years (range: 59-94 months). They had already started to read and had developed some phonological awareness skills. A relationship between speechreading and reading independent of phonological awareness may have existed in this group at a previous timepoint but was not evident at the time the children were tested. In addition, despite the full mediation observed in the model it is possible that the direct pathway between speechreading and reading was not maintained for power reasons (Rucker et al., 2011) as discussed below. However, Chapter 8 showed no evidence to support an effect of speechreading training on reading via a route independent of phonological awareness.

Turning to the data from hearing children, the model in Chapter 7 suggests that phonological awareness fully mediates the relationship between speechreading and reading just as it does in deaf children. As discussed in Chapter 2, although the term phonological representations is generally used to refer to the ability to represent sound patterns within a spoken language it is likely that these representations are multimodal. There are multiple sources of information that may contribute to the representation of speech, including the auditory speech information, visual speech information, orthography and the person's own articulation (Carroll et al., 2003; E. A. Elliott et al., 2011; Jerger, Tye-Murray, & Abdi, 2009; Leybaert, 1993; Patterson & Werker, 2003). In a hearing child it is likely that the representations are weighted towards auditory speech information. However, previous studies have shown that infants as young as 2 months old are able to match auditory and visual speech information (Patterson & Werker, 2003). This suggests that even at a young age, hearing infants are able to represent and integrate both auditory and visual speech information. In hearing adults, visual speech information is known to improve speech perception in noisy environments (Lusk & Mitchel, 2016; Mitchel & Weiss, 2014; Sumby & Pollack, 1954) suggesting that having a representation of multiple sources of speech information makes speech perception more robust to disruption in one stream of information. The model in Chapter 7 is consistent with the idea that phonological representations are multimodal, representing visual speech information as well as auditory speech information in young hearing children.

Hearing adults and children who tend to have poorer phonological awareness skills, for example those with developmental dyslexia or developmental language disorder (DLD), also show poorer visual speech perception (Heikkilä, Lonka, Ahola, Meronen, & Tiippana, 2016; Knowland, Evans, Snell, & Rosen, 2016; Mohammed et al., 2006). Chapter 7 found that speechreading skill correlates with phonological awareness in young typically developing hearing children (r = .47). This is in contrast to Harris et al. (2017) who found a much smaller relationship between speechreading and phonological awareness in 5-to-6-year-old hearing children (r = .11). One explanation for the difference in results is that speechreading and phonological awareness were measured with multiple-choice tasks in Harris et al.'s (2017) study but with free-response tasks in Chapter 7. Free responses allow for more variation and tend to be more reliable and sensitive measurements.

Although a full mediation between speechreading and reading via phonological awareness was observed for both deaf and hearing children in the models in Chapters 6 and 7 this does not preclude the possibility that other variables contribute to this relationship. Rucker, Preacher, Tormala, & Petty (2011) highlighted that in a mediation analysis if an indirect effect is significant (speechreading -> phonological awareness -> reading) this suggests that this pathway is a good, although not necessarily the only, explanation of the data. A full mediation can occur for a number of reasons, including low power for the analysis. As the sample was relatively small for structural equation modelling with deaf children (n = 66), low power may explain the full rather than partial mediation observed in that group at least. Therefore, it is possible that there are other factors, such as vocabulary and attentional skills that may also mediate the relationship between speechreading and reading.

9.3 What is the causal direction of any relationships between speechreading, phonological awareness and reading?

The following sections will address three possible causal directions of the relationship between speechreading, phonological awareness and reading in light of the studies reported in Chapters 6-8. These are: 1) that speechreading improves phonological awareness ability and consequently reading ability; 2) that phonological awareness is a causal influence on both speechreading and reading; and 3) that the direction of the relationship between these factors may change across development.

9.3.1 Speechreading improves phonological awareness ability and consequently reading ability

The working hypothesis presented throughout this thesis has been that speechreading and reading in deaf and hearing children may be related because speechreading provides one of the sources of information for developing phonological representations (Kyle & Harris, 2011; Mohammed et al., 2006). This is especially the case for deaf children with several studies demonstrating the use of speechread information during phonological matching tasks (Dodd & Hermelin, 1977), linguistic serial recall tasks (Dodd et al., 1983) and in their spelling errors (Burden & Campbell, 1994; Hanson et al., 1983; Leybaert & Alegria, 1995; Sutcliffe et al., 1999). In addition, previous studies have found that deaf children's speechreading skill relates to performance on phonological awareness tasks, both concurrently (Kyle & Harris, 2006) and longitudinally (Kyle & Harris, 2010). The results from Chapter 7 show that there is also a concurrent relationship in hearing children.

The models in Chapters 6 and 7 were theoretically motivated by this working hypothesis. However, as both models were based on concurrent data, the direction of causality cannot be determined. One way to establish whether visual speech perception contributes to the development of hearing children's phonological awareness and reading skills is to train speechreading. Whether or not speechreading skill is fixed or can be improved has been debated (Bernstein et al., 2001; Blumsack et al., 2007; Dodd et al., 1989; Gagne & Dinon, 1991; Gesi et al., 1992; Massaro et al., 1993; Montgomery & Sylvester, 1984; Summerfield, 1992; Walden et al., 1981, 1977). Chapter 8 trained speechreading in hearing children to test the hypothesis that any observed improvements in speechreading may lead to gains in phonological awareness and reading. Young hearing children were trained on the computerised Speechreading Training and Reading (STAR) programme for 10 minutes a day, 5 days a week for 3 weeks. The children who undertook the speechreading training were significantly better on a single word speechreading task at the immediate and three-month post-test than their classmates who had not done the intervention. However, there was no improvement on phonological awareness as a result of the intervention except in the poorest performing children. There were also no effects on reading.

The lack of strong evidence for a causal role for visual speech information in the development of phonological awareness and reading in the intervention study in Chapter 8 may be for methodological reasons. First, all of the hearing children were in their first year at school (Reception). In the UK there is a very strong focus on phonics in the early school years. Therefore, the children made substantial gains on the phonological awareness tasks and reading from pre-test to post-test (across 5 weeks starting in January or

February). In this context, any improvement as a result of the speechreading training may have been too small to be detected. In Reception the children receive 30 minutes of phonics training every day. Therefore, the 10 minutes of speechreading training that the intervention group received each day may have been insufficient in the context of the phonics training both groups received to observe any differences from the control group in their phonological awareness skills. One way to address this issue would be to train speechreading in pre-school children before they receive phonics training in school. In the training study in Chapter 8 Reception children in their second term (January-March) at school were chosen as participants as piloting in October (one month after the start of school) had shown that younger children were not able to complete the STAR games or the phonological awareness or reading tasks.

There were no group-level effects on the blending task as a result of the speechreading intervention, despite the fact that the intervention included silent blending training. However, those in the intervention group who started with low phoneme blending scores showed greater improvement on the phoneme blending task than those in the control group with low initial scores. As discussed in Chapter 8 this interaction may be explained by a ceiling effect on the phoneme blending task. It is possible that the ceiling effect masked a main effect of the intervention, as children who scored at the top end of the task (n = 14 items) could not improve any further on the task. Therefore, it is possible that speechreading training could play a causal role in phonological awareness ability that was not identified by the intervention study in Chapter 8. An alternative explanation is that as there were few children scoring at the lower end on the phoneme blending measure this led to an exaggeration of the differences between groups in post-test scores for the lowest performing children. However, it is likely to be specifically these children with poorer phonological awareness skills who may benefit from emphasising visual speech information in order to support their understanding of the sublexical structure of speech. Therefore, it is possible that visual speech information contributes to phonological awareness ability in hearing children but that this effect is of most relevance to the poorest performing children. Future investigations into the role of visual speech perception in phonological awareness and reading ability should consider targeting children who are struggling with these skills (see Knowland et al., 2016).

Another explanation for the lack of evidence for a causal relationship between speechreading and phonological awareness is that speechreading may have improved the children's phonological representations, but that the improvement was not sufficient to boost the children's ability to *manipulate* those representations. Manipulation is precisely what was measured in our phonological awareness outcome measures. The phoneme deletion task is a strong predictor of reading ability in young hearing children (Hulme et al., 2009). However, it is also a fairly abstract and complex task for young children to complete, requiring manipulations and skills that are unlikely to be improved by speechreading skills, such as memory and attention. Therefore, improvements in phonological representations as a result of speechreading training may not have translated into improvements in phoneme deletion. Phonological representations cannot be directly measured. One way to get an indication of the quality of phonological representations is to measure speech production, provided there are no physical reasons for poor articulation (Carroll et al., 2003; Stackhouse & Wells, 1997). In the intervention in Chapter 8 the children's speech production during the vocabulary task at pre-test and post-test was videoed to allow for future analysis of changes in speech production as a result of the intervention. The video data collected are now being used by an MSc Speech and Language Sciences student, who will score audio-visual speech production, as an indication of phonological representations (as in the STAR_D project). In the STAR_D project, visual and auditory speech output was scored at pre-test and post-test as a measure of phonological representations in those young deaf children. The STAR_D project showed that the poorest performing children improved on their speech output scores at the three-month post-test as a result of the speechreading intervention (Pimperton et al., submitted). This result was used to suggest that despite no differences between the intervention and control groups on an explicit phonological awareness measure at post-test, the deaf children who took part in the STAR intervention benefitted from improved phonological representations. Therefore, analysis of the video data from the current study will help to determine whether the same is true for the hearing children involved in the STAR intervention in Chapter 8.

9.3.2 Phonological awareness is a causal influence on both speechreading and reading

Another explanation of the patterns observed in the structural equation models in Chapters 6 and 7 is that phonological awareness is a causal influence on *both* speechreading and reading ability. It may be that having better phonological awareness both facilitates speechreading and reading ability, explaining the relationship between them. For hearing children, phonological representations and awareness may be primarily developed through auditory speech perception. They may then use this knowledge to aid their speechreading ability, thus explaining the relationship between speechreading and reading in hearing children. However, they must be able to integrate auditory and visual speech information (e.g. Kuhl & Meltzoff, 1982; Patterson & Werker, 2003) in order to use phonological representations based on auditory speech perception to support speechreading. Therefore, it is likely that both visual speech perception and auditory speech perception contribute to phonological representations. As a result, even if better phonological awareness leads to better speechreading ability, this relationship is likely to be reciprocal.

If phonological awareness has direct effects on both speechreading and reading, with better phonological awareness skills predicting better speechreading skills, we might expect hearing children to have better speechreading skills than their deaf peers. Deaf and hearing children have been showed to not differ on their speechreading ability (Chapter 5; Kyle et al., 2013). Therefore, even if phonological awareness skills do contribute to speechreading ability, this is unlikely to be the only explanation for the correlation between speechreading and phonological awareness observed in Chapters 6 and 7.

For deaf children, a direct relationship between phonological awareness and both reading and speechreading is unlikely to explain the relationship between these variables found in Chapter 6. Although any aided or residual auditory information may contribute to the development of phonological representations, visual speech information is likely to be more heavily weighted as a source of phonological information. Previous studies have shown that deaf children use speechread information during phonological awareness tasks (Dodd & Hermelin, 1977; Dodd et al., 1983) and their spelling errors show an influence of speechread information (Burden & Campbell, 1994; Hanson et al., 1983; Leybaert & Alegria, 1995; Sutcliffe et al., 1999). In addition, deaf children's speechreading skills correlate with their phonological awareness both concurrently (Kyle & Harris, 2006) and longitudinally (Kyle & Harris, 2010). The STAR_D project found that for the poorest performing children training speechreading led to improvement in speech production measures, used as an indication of the underlying phonological representations. Therefore, it is unlikely that in young deaf children phonological awareness is simply a direct causal influence on both speechreading and reading ability.

Although speechreading is likely to play a causal role in the development of phonological awareness in deaf children, it is also possible that deaf children develop their phonological awareness skills through reading. As highlighted previously in this Discussion, Kyle (2015) suggested that the relationship between phonological awareness and reading does not emerge until after children have started to read. This is derived from studies showing that early phonological awareness in deaf children does not predict later reading ability, but instead early reading skills predict later phonological awareness (Harris & Moreno, 2004; Harris et al., 2017b; Kyle & Harris, 2010). In the model in Chapter 6 the deaf children were very variable in their reading ability but had already started to read. Therefore, it is possible that in the concurrent model in Chapter 6 phonological awareness skills had already been established to some extent through orthography. However, Kyle (2015) suggested that the speechreading-reading relationship precedes the influence of reading on phonological awareness. Therefore, although better phonological awareness may lead to better speechreading, this is unlikely to be the only direction of the relationship between these two factors.

9.3.3 Changing developmental patterns

The previous sections outline two possible directions for the relationship between speechreading and phonological awareness in deaf and hearing children. However, it is likely that speechreading, phonological awareness and reading are all reciprocally related to each other and that the direction of the relationships between these three factors changes throughout development. Previous researchers have suggested changing relationships between speechreading, phonological awareness and reading in deaf children. For example, Kyle (2015) suggested that once phonological awareness skills have been established they then form a reciprocal relationship with reading skills in deaf children. In addition, some studies have found that early phonological awareness does not predict later speechreading ability (Harris et al., 2017; Kyle and Harris, 2011) in deaf children. However, Kyle and Harris (2010) showed that initial phonological awareness skills at 7 years old did not predict speechreading skill a year later, but phonological awareness skills at 8 years old did predict speechreading skill two years after that. This supports the idea that the relationship between speechreading and phonological awareness may change across development in deaf children.

With regard to hearing children, the evidence reviewed in Chapter 7 suggests that visual speech perception relates to the discrimination and identification of phonemes in infancy (Weikum et al., 2007). Being able to discriminate between phonemes indicates that these infants are developing well-defined phonological representations. This supports the idea that in the earliest stages of phonological development visual speech information is important. Having well-defined phonological representations will in turn enable children to develop their phonological awareness skills. There also appears to be a reciprocal relationship between phonological awareness and reading development. Early phonological awareness skills predict later reading ability (Melby-Lervåg, Lyster, et al., 2012), and in turn learning to read appears to improve phonological awareness skills (e.g. Castles & Coltheart, 2004; Nation & Hulme, 2011), as discussed in Chapter 2. Finally, it may be that having strong multimodal phonological representations and good phonological awareness (through auditory speech, visual speech and reading) lead to better speechreading.

Future studies should investigate how the relationship between speechreading, phonological awareness and reading changes through development for both deaf and hearing children. An important way to understand the developmental relationships between speechreading, phonological representations and awareness, and reading ability is to use longitudinal analyses. Snowling, Lervåg, Nash and Hulme (2018) found that categorical perception skills related to phonological awareness and reading concurrently but that phonological awareness did not mediate the relationship between categorical perception and reading in a longitudinal dataset. It may be possible in the future to use the STAR_D and STAR_H data to model the longitudinal relationships between speechreading, phonological awareness and reading in young deaf and hearing children. For example, growth curve modelling could be used to examine whether early speechreading skill predicts the developmental trajectory of phonological awareness and reading development in deaf and hearing children rather than just whether early skills predict later outcomes. In addition, the mediating role of phonological awareness in the relationship between speechreading and reading can be modelled longitudinally in order to investigate the direction of the relationship.

9.4 Summary and implications

This thesis provides evidence for a triangular relationship between speechreading, phonological awareness and reading in both deaf and hearing children. Further research is required to establish the direction of the relationships between these three factors and what other factors may be involved in the relationship between speechreading and reading. The similarity in results between deaf and hearing children in each study described in this thesis is striking. The results suggest that deaf and hearing children access visual speech information in a similar way and that it also relates to reading ability in a similar way in these two groups. The work in this thesis has several implications for literacy development in both deaf and hearing children.

9.4.1 Deaf children

Auditory verbal therapy (AVT, www.avuk.org) is an early intervention for deaf children which focuses on auditory information for language development, discouraging use of visual cues. The studies in this thesis provide several challenges to the use of AVT for deaf children's language and literacy development. First, Chapter 5 showed that both deaf and hearing children attend to the mouth when speechreading and that the extent to which they look at the mouth during speechreading correlates with their speechreading ability. This suggests that being able to see a speaker's mouth when they are speaking facilitates speechreading. As deaf children have reduced access to auditory speech information, even with a hearing aid or cochlear implant, vision is an important source of speech information. This is in line with previous literature showing that visual speech information can boost auditory speech perception in noise for hearing adults

(Lusk & Mitchel, 2016; Mitchel & Weiss, 2014; Sumby & Pollack, 1954). In addition, work with hearing infants suggests that visual speech information can boost phonetic discrimination (Erdener & Burnham, 2013; Weikum et al., 2007) and that infants can match information across modalities (Patterson & Werker, 2003). Therefore being able to access information from the mouth during speech perception is likely to aid the formation of robust multimodal phonological representations.

The studies in Chapters 6 and 7 show that speechreading ability relates to phonological awareness and reading ability in deaf and hearing children. Further work with the video data collected in Chapter 8 is needed to establish whether better speechreading leads to better phonological representations. However, the relationship between speechreading, phonological awareness and reading suggests that even if some time is spent emphasising the auditory speech cues in early interventions, visual speech perception may be another factor to consider and utilise in interventions. The evidence presented here suggests that practices that discourage a child's use of visual information during speech perception are potentially restricting the child's ability to form robust multimodal phonological representations and may have adverse consequences for the development of their phonological awareness and reading skills. Long-term intervention studies are needed to test these ideas.

9.4.2 Hearing children

For hearing children the studies in this thesis suggest a potential role for visual speech perception in the development of phonological awareness and reading. The interaction in Chapter 8 suggests that speechreading may play a causal role in the development of phonological awareness in those children with poorer phonological awareness. This supports the idea that phonological representations are multimodal and that visual speech information may facilitate the development of phonological representations regardless of whether a child can hear or not. As argued in Chapter 2, it is likely that in hearing children the weighting of the information is likely to be biased towards what they can hear but that visual speech information also relates to phonological representations. It is possible that emphasis on visual speech information may help hearing children with poorer phonological awareness to develop their phonological awareness skills by providing an additional source of information about the sublexical structure of speech. Further work targeting those young hearing children with poor phonological awareness will help to clarify the relationship between visual speech perception and phonological awareness.

Appendix A

Computerised speechreading training for deaf children: A randomised controlled trial

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Abbreviated title: Computerised speechreading training for deaf children: An RCT

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Abstract

Background

The majority of children born deaf find learning to read a very difficult task. Previous longitudinal studies have demonstrated that speechreading (lipreading) is a good predictor of reading outcomes in young deaf children. It has been proposed that speechreading contributes to a deaf child's sublexical (phonological) representations of spoken words and that these representations can then support early word reading, in a similar way to that observed in hearing children. Here we tested this model by developing and assessing the effects of a computerised speechreading training programme on speechreading, phonological and reading skills.

Methods

Sixty-six deaf 5-7 year olds were randomised into two arms: speechreading training and maths training. The maths training gaming environment paralleled the speechreading training. Both training programmes involved 10 minutes training a day, 4 days a week, for 12 weeks. Children were tested by blinded assessors on a battery of assessments before training (T1), immediately following training (T2), 3 months after completing training (T3) and 11 months after completing training (T4). This study was pre-registered on the Open Science Framework https://osf.io/ygz7f/.

Results

We found significantly greater gains in speechreading performance, on tasks using both trained and untrained stimuli, in deaf children who carried out the speechreading training compared those who completed the maths training. There was also some evidence of beneficial effects of the speechreading training on phonological skills. However, these effects were weaker and no benefits were seen to word reading.

Conclusions

Speechreading skill is trainable in young deaf children. However, gains in speechreading following training are not necessarily sufficient to impact word reading outcomes. Speechreading training may be more effective at supporting reading when fully embedded in a broader literacy programme. Nevertheless, a training game that can improve speechreading is likely to be of great interest to professionals working with deaf children.

Keywords: deaf; speechreading; lipreading; reading; phonological awareness; intervention; training; RCT

Abbreviations: ToCS (Test of Child Speechreading); RCT (Randomised Controlled Trial)

Introduction

Speechreading refers to the ability to understand speech solely on the basis of visual, rather than auditory, perceptual information. More commonly known as lipreading, the term speechreading acknowledges the fact that there is more to understanding visual speech than solely what is seen on the lips (Arnold, 1997). For many profoundly deaf children, speechreading provides their main access to spoken language. For others, visual speech information can support speech perception by complementing impoverished auditory speech information provided via cochlear implants or digital hearing aids.

Given that speechreading provides visual access to spoken language, it is perhaps not surprising that speechreading has been argued to play an important role in deaf children's reading development (Kyle et al., 2006; 2010). Support for this comes from cross-sectional studies that have demonstrated concurrent correlations between speechreading and reading abilities in deaf children (Kyle et al., 2006; Kyle et al., 2016) and adult readers of both English (Mohammed, Campbell, MacSweeney, Barry, & Coleman, 2006) and Spanish (Rodriguez-Ortiz et al., 2017). Furthermore, longitudinal studies have documented predictive relationships between early speechreading skills and later reading outcomes in young deaf children

(Kyle et al., 2010; 2011; Harris et al., 2017); better speechreading skills are associated with better subsequent reading outcomes. Data from the Kyle et al (2010) longitudinal study further suggested that this relationship between speechreading and reading is mediated by phonological processing in deaf children. Information about the sublexical structure of speech derived from speechreading may contribute to the formation of phonological representations of spoken language in deaf children, which they can then bring to the task of learning to read. This is important since the vast majority of deaf children find learning to read to be a difficult task, with many studies reporting significantly poorer reading skills in deaf children than their hearing peers (Wauters, et al., 2006; Qi & Mitchell, 2011). Here we report a study in which we tested whether we could train speechreading skills in young deaf children. We also tested the above model by assessing the impact of this training on phonological and single word reading skills.

Whether it is possible to train speechreading skills has been the focus of debate following discrepant findings. There are very few studies of speechreading training with deaf adults. Where speechreading gains have been reported following speechreading training, in contrast to a control group, these have often been small (e.g., Bernstein Auer & Tucker, 2001). Even less published evidence is available regarding speechreading training in deaf children, despite researchers highlighting its potential benefits

(Arnold, 1993). Van Uden (1983) tested profoundly deaf 8-14 year olds on a mixed speechreading/ articulation programme. Participants who viewed themselves producing speech made greater speechreading gains than groups that viewed a teacher producing speech.

Despite the paucity of high quality evidence from speechreading training studies, numerous studies demonstrate a speechreading advantage for adults who have experienced congenital or early onset deafness (Auer & Bernstein, 2007; Bernstein, Demorest, & Tucker, 2000; Mohammed et al., 2006; Pimperton et al; 2017). This suggests that increased experience of, and attention to, visual speech early on in life can result in a form of perceptual compensation and bring about improvements to visual-only speech perception. This is consistent with studies in other modalities which have indicated enhanced perceptual compensation at earlier ages (e.g., Gougoux et al. (2004). The responsivity of speechreading skill to early environmental experience supports the hypothesis that it may be amenable to training in young children.

In summary, evidence from longitudinal studies of deaf children's reading development indicates that good speechreading skills may support the process of learning to read in deaf children. This raises the possibility that training speechreading skills in deaf children, if effective, could bring benefits to their reading development. In the present study we created, and evaluated in a Randomised Controlled Trial, a 12 week computer-based adaptive speechreading intervention for 5-7 year old deaf children. This allowed us to test two key hypotheses:

1. That speechreading skills in young deaf children can be improved by training.
2. That improvements in speechreading will transfer to improvements in phonological and reading skills.

Methods

Design

A single blind randomised controlled trial was conducted with deaf children aged 5-7 years old. Children were tested on an assessment battery prior to training (T1), and then randomised to complete either speechreading or maths (control) training. Follow up assessments were conducted immediately following the completion of the intervention (T2) and three months later (T3). The study design, analysis plan and sample size calculations were pre-registered on the Open Science Framework (https://osf.io/ygz7f/). Following data collection at T3 it was decided to test the children again 11 months after the completion of the intervention (T4) to examine the durability of any intervention effects. Although not registered in the OSF study design, these data are also reported here for completeness.

Ethical approval for the study was provided by the UCL Research Ethics Committee.

Participants

Schools for deaf children, mainstream schools with hearing impaired units (HIUs) and local authority support services for deaf children were asked to identify children who met the following eligibility criteria:

1. Aged between 5 and 7 years at the time of the first assessment.

2. With a severe or profound bilateral hearing loss which had onset before the age of 12 months.

3. Able to meet the physical and attentional demands of playing a computer game for 10 minutes a day.

The caregivers of 70 deaf children provided informed consent for their child to participate in the trial.

The CONSORT diagram in Figure 1 shows the flow of participants through the trial. See Table S1 [*not included in this thesis*] for the CONSORT checklist. Four participants were excluded prior to randomisation. One had insufficient functional language skills to complete any assessments, two did not meet the audiological inclusion criteria and one did not meet the age inclusion criteria. The remaining 66 participants were randomised into the two arms of the trial (intervention = 33; control = 33). Group allocation was conducted independently by the University of York Trials Unit, using minimisation (1:1 allocation ratio) on the following criteria:

1. Total Test of Child Speechreading score at T1 (above vs. below a median split)
2. Communication preference (oral vs. sign or speech with sign)
3. Year group ($<$Y2 vs. $\geq$ Y2)

There were no significant differences between the intervention and control groups on demographic, audiological and educational factors (see Table 1). Two children (one intervention, one control) were lost to follow-up (see Figure 1) and did not provide assessment data at T2 and T3. A further 2 children, both in the active control group, were lost to follow up at T4. The researchers carrying out the assessments of the participants on the study outcomes were blind to the group allocation of the participants.

Figure 1: Flowchart documenting movement of participants through the phases of the trial.

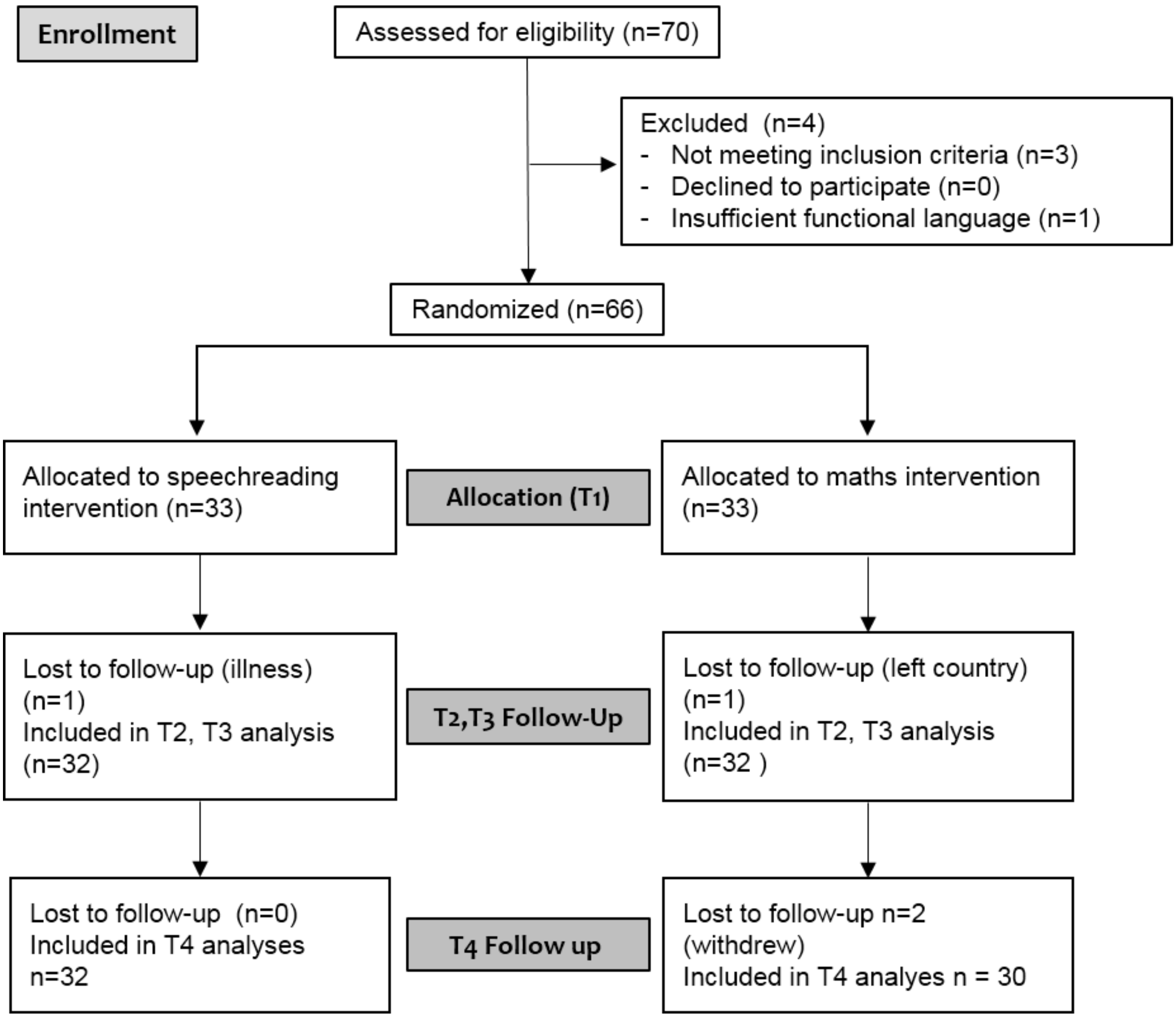

Table 1. Participant characteristics of the intervention and control groups at baseline (T1)

	Intervention (*N* = 33)	Active control (*N* = 33)	F (*p*)
Chronological age (months) Mean (*SD*), Range	73.24 (8.08), 61 - 94	71.94 (7.68), 59 - 91	0.45 (.50)
Non-verbal ability (raw score) Mean (*SD*), Range	6.24 (2.29), 2 - 14	6.82 (2.63), 1 - 14	0.90 (.35)
			X^2 (*p*)
Year Group (%)			
< Y2	55	58	0.06 (.80)
≥ Y2	45	42	
Communication preference (%)			
Spoken English only	30	30	0.00 (1.0)
Sign or sign with speech	70	70	
School Setting (%)			
School for deaf children	18	21	0.10 (.95)
Hearing impaired unit	61	58	
Mainstream school	21	21	
Device Use (%)			
No device	6	6	3.11 (.37)
Bilateral CIs	48	39	
One HA, one CI	6	0	
Bilateral HAs	39	55	
Unaided Severity Category (%)			
Severe*	42	33	0.58 (.45)
Profound	58	67	

* Five children (two in the speechreading group, three in the maths group) had a hearing loss that at their latest hearing assessment was in the moderate category in their better ear, but severe or profound in the contralateral ear, these children were included in the severe category.

Interventions

Both the speechreading and maths interventions were run within a suite of seven space-themed computer games (see Figure S1 for examples). Adaptive algorithms were established to enable a child to progress through the training at a pace appropriate to their ability. The games were designed to run across 48 ten-minute sessions. Each ten-minute session was packaged so that there was a narrative structure to the space games and the child received a virtual reward at the end of each session to collect in an online 'trophy cabinet'. Full details about both interventions can be found in the online Supplementary Methods.

In-game assessments and assessments at pre-test (T1), post-test (T2), after intervention follow up at 3 months (T3) and 11 months (T4)

Below is a summary of assessments. More detail of each assessment is provided in the online Supplementary Methods.

In-game assessments. Within both the speechreading and maths intervention programmes children completed an assessment of their speechreading skills every eight training sessions. In each assessment trial the children viewed a video of an unknown talker saying one of the trained words and had to choose the corresponding picture from a choice of four.

Speechreading

The pre-specified *primary outcome measure* in this study was total standard score on the Test of Child Speechreading (ToCS; Kyle, Campbell, Mohammed, Coleman, & MacSweeney, 2013). The ToCS is a standardised computerised assessment of speechreading ability that comprises three subtests; single words, sentences and short stories. In all tasks the participant is required to match the spoken target to a picture. At T4, children only completed the ToCS single word subtest due to the difficulty of the sentence and story subtests for these young children.

The following pre-specified *secondary outcome measures* were also collected.

Speechreading: Test of Child Speechreading (ToCS) – Everyday Questions test

The children completed the Everyday Questions Subtest from the ToCs. This required them to watch silent videos (n=12) of two talkers saying questions they might encounter in everyday life (e.g. where do you live?) and tell the experimenter what they thought the question was.

Vocabulary

A naming task, using the pictures from the training (N=74), was used to assess participants' knowledge of the vocabulary used in the speechreading training.

Audio-visual speech production (AV speech production)

Videos of children naming pictures for the vocabulary assessment were used to establish measure of the quality of their audio-visual speech production was derived from 30 of the trained words. This measure provided an indirect measure of the quality of the child's phonological representations (Stackhouse and Wells, 1997).

Phonological awareness

A novel phonological awareness task, based on that of Kyle and Harris (2006), was developed using stimuli from the speechreading training to assess the children's awareness of spoken English phonology at the level of onset and rime.

Letter-sound knowledge

The Letter-Sound Knowledge subtest of the York Assessment of Reading for Comprehension Primary School Edition (YARC; Snowling et al., 2009) was used to assess children's knowledge of the correspondence between letters and sounds.

Word reading

The Early Word Recognition Test and the Single Word Reading Tests from the YARC (Snowling et al., 2009) were used. In addition, we developed a test to assess single word reading for stimuli included in the speechreading training (n=24 trials). A reading composite score was created by summing each child's z scores on the three word reading measures.

Number skills

Three measures of number skills were administered. 1) The Early Number Concepts section of the BAS-III (Elliot & Smith, 2011) 2) Test of Basic Arithmetic and Numeracy Skills (Hulme, Brigstocke, & Moll, 2016). 3) Children were asked to count to 30, with the highest number they could reach being their score on this task. A Number Skills composite score was created by summing each child's z scores on the three measures of number skills. At T4 only the measure of addition and subtraction fluency was administered.

Statistical methods

Differences between the intervention and control groups on the outcome variables at T2, T3, and T4 were tested using ANCOVAs with the outcome variable at T2, T3, or T4 as the dependent variable, performance on the same variable at baseline (i.e. T1) as a covariate, and group (i.e. intervention vs. control) as a fixed factor. In those cases where the residuals of the ANCOVA model showed a significant deviation from the normal distribution, ANCOVA models with bootstrapped SEs (1000 bootstrap samples) were run. For all ANCOVA models, equality of slopes was assessed by including the interaction between covariate and group. Cohen's *d* provided a measure of the intervention effect size and was calculated by dividing the difference in progress between the intervention and control groups by the pooled initial *SD* (Morris, 2008).

Results

Assessment data were collected from 32 children in each group at T1, T2 and T3. At T4 data were collected from 32 children in the speechreading group and 30 children in the control group.

The mean time between the first two assessment points (T1 and T2) was 5.42 months (*SD*=1.21). The wide range within the whole sample (3-8 months) was a result of school logistical constraints and child illness. However, there were no significant differences between the intervention and control groups in terms of their T1-T2 distance (Speechreading: M=5.34, *SD*=1.18, range=3-7; Maths: *M*=5.50, *SD*=1.24, range=3-8; $t(62)=0.52$, $p=.61$, bootstrapped BCA 95% CI=–0.76, 0.42). The distance between T2 and T3 in the whole sample was less variable (*M*=2.66, *SD*=0.48, range=2-3 months) and there were no significant differences between the intervention and control groups (Speechreading: *M*=2.69, *SD*=0.47, range=2-3; Maths: *M*=2.63, *SD*=0.49, range=2-3; *t*(62)=0.52, *p*=.61, bootstrapped BCA 95% CI=–0.17, 0.31). The distance between the T3 and T4 assessments averaged 7.92 months (*SD*=0.91, range=6-10 months) and did not differ significantly between the intervention (*M*=7.94, *SD*=0.98, range=6-10) and control (*M*=7.90, *SD*=0.84, range=6-10) participants (*t*(60)=0.16, *p*=0.87, bootstrapped BCA 95% CI=-0.43, 0.48).

Adherence to intervention

There was substantial variation in the number of training sessions completed by children in the speechreading training group (*M*=36.77, *SD*=16.88, range=0 - 48). This was due to school logistical and technological constraints and child illness. There were no significant differences between the two groups in total number of intervention sessions completed (Speechreading: *M*=35.25, *SD*=18.61; Maths: *M*=38.28, *SD*=15.11; *t*(62)=0.72, *p*=.48, *d*=0.18). Six children did not complete any sessions but were still included in the intention to treat analyses. Very similar numbers of children completed all 48 training sessions in the speechreading and math training groups (Speechreading = 18; Maths = 19).

Group comparisons on outcome measures – Intention to Treat analyses

Descriptive statistics, including means and *SD*s, for the performance of all participants on the outcome measures at Baseline (T1), Immediate Follow-up (T2), Delayed Follow-up (T3) are presented in Table 2 and for the second Delayed Follow-up (T4) in Table S2 (due to different number of participants and minor changes in measures at T4). Also presented are Cohen's *d* effect sizes and results of the ANCOVAs comparing the two groups on each outcome while adjusting for their baseline performance.

Table 2. Intention to treat analyses: Means and *SD*s, for all participants on the outcome measures at Baseline (T1), Immediate Follow-up (T2) and Delayed Follow-up (T3). Also presented are Cohen's *d* effect sizes and results of the ANCOVAs comparing the two groups on each outcome, at T2 and T3, while adjusting for their baseline performance.

	Intervention (N = 32)		Active control (N = 32)		Cohen's d^{+}	β (*p*) [95% CI]
	M	*SD*	*M*	*SD*		
ToCS Total Standard Score						
T1	94.78	10.96	95.84	13.48		
T2	93.53	11.14	94.13	12.48	0.04	0.02 (.99) [-4.76, 4.80]
T3	95.16	11.88	95.03	12.79	0.10	0.78 (.75) [-4.12, 5.69]
ToCS Everyday Questions						
Words identified (max = 62)						
T1	5.50	9.27	5.53	11.13		

T2	10.41	10.58	8.97	12.79	0.14	1.47 (.43) [-2.34, 4.94][1]
T3	14.16	12.40	10.34	14.70	0.38	3.84 (.09) [–.42, 8.52][1]
Items correct gist (max = 12)						
T1						
T2	0.41	0.95	0.75	1.98		
T3	1.28	1.73	1.06	2.03	0.38	0.49 (.17) [-0.24, 1.14][1]
	1.78	2.28	1.22	2.28	0.61	0.91 (.03) [0.17, 1.65][1]
Vocabulary (max = 74)						
Overall						
T1	54.50	5.78	53.78	9.33		
T2	63.66	6.56	60.69	8.76	0.30	2.45 (.07) [-0.23, 5.13]
T3	63.59	7.31	62.81	8.26	0.008	0.32 (.83) [-2.71, 3.35]
Spoken						
T1	41.78	17.15	37.22	22.33		
T2	53.00	20.38	44.63	23.70	0.19	3.58 (.06) [-0.25, 7.22][1]
T3	54.69	18.84	49.16	22.59	0.05	1.07 (.57) [-2.63, 4.76]
AV Speech Production (%)						
Updating[2]	65.22	31.67	56.23	31.23		
T1	69.80	28.65	58.94	33.24	0.06	2.24 (.31) [-1.17, 5.92][1]
T2	72.14	30.20	60.15	33.67	0.10	3.28 (.23) [-1.79, 9.03][1]
T3						
Phonological Awareness (max = 24)						
T1	10.28	4.23	10.41	5.28		
T2	12.88	5.53	12.16	6.07	0.18	0.84 (.36) [-0.96, 2.64]
T3	14.31	5.83	13.03	5.92	0.30	1.39 (.19) [-0.71, 3.49]
Letter-sound Knowledge (max = 17)						
T1	11.34	4.99	10.47	5.70		
T2	13.03	5.14	12.03	6.02	0.02	0.16 (.78) [-0.88, 1.30][1]
T3	13.31	5.53	11.81	6.20	0.12	0.63 (.33) [-0.58, 1.93][1]
Word Reading (Z-score composite)						
T1	0.01	2.89	-0.01	2.90		
T2	0.12	2.73	-0.12	3.02	0.08	0.23 (.36) [-0.28, 0.72][1]
T3	0.12	2.71	-0.12	2.99	0.08	0.24 (.48) [-0.43, 0.95][1]
Number Skills (Z-score composite)						
T1	0.08	2.54	-0.08	2.54		
T2	0.02	2.51	-0.02	2.58	-0.06	-0.10 (.76) [-0.77, 0.56]
T3	0.001	2.35	-0.001	2.66	-0.06	-0.14 (.69) [-0.81, 0.54]

[+]Cohen's *d*: Difference in progress between groups divided by pooled initial *SD*

[1]ANCOVA models run with bootstrapped SEs, bootstrapped bias-corrected and accelerated 95% CIs are reported

[2]N = 30 in each group

There was no significant difference between the intervention and control groups on our pre-specified primary outcome variable, total ToCS standard score, at either T2 (d=0.04) or T3 (d=0.10) nor on the ToCS word subtest at T4 (d=0.14). Of the pre-specified secondary outcome variables, there was evidence of an effect of speechreading training on the ToCS Everyday Questions task after adjusting for performance at T1. This was greater for the 'Items correct gist' measure than the 'Words identified' measure. There was a moderate effect for 'Items correct gist' at T2 (d_{Items}=0.38; d_{Words}=0.14) and a larger, significant, effect for the same measure at T3 (d_{Items}=0.61; d_{Words}=0.38). The training effect was smaller at T4, 11 months after training ended (d_{Items} =0.22; d_{Words}=0.31), and was no longer significant.

There was a moderate but non-significant effect of speechreading training on phonological awareness at T3 (d=0.30) which was small at T2 (d=0.18) and T4 (d=0.17). There were small to moderate effects of training at T2 on overall vocabulary (d=0.30) and spoken vocabulary (d=0.19), with no evidence of sustained effects at T3 ($d<0.01$ and d=0.05 respectively) or T4 (d=-0.12 and d=–.08 respectively). The effect of training on word reading was small and non-significant at T4 (d=0.22) and T2 and T3 (ds=.08). Finally, there was no evidence of an effect of intervention group on Letter-Sound Knowledge (T2/T3) or Number Skills (T2/T3/T4) (ds: -0.06 – 0.12).

Only the ANCOVA model for AV speech production at T2 did not meet the assumption of equal slopes. The interaction between intervention group and the covariate (T1 audio-visual speech production) was significant (unstandardized slope = 0.18 [95% CI .07, .29], p=.003) indicating a shallower slope in the intervention group than the maths control group. This pattern indicated that the speechreading intervention was more effective for children starting with lower scores on this measure. Follow up tests showed that the groups did not differ at post-test at the mean of the covariate ($F(1, 56)$=1.19; p=.280). However, for children scoring at 1 standard deviation below the mean of the covariate, there was a significant advantage for the intervention group ($F(1, 56)$=9.57; p=.003). This pattern needs to be interpreted with caution but suggests that speechreading training was effective for children in improving T2 speech output in children who started with particularly low scores on this measure. A similar pattern was seen for the same variable at T3 though the interaction term between intervention group and the covariate was not significant (unstandardized slope = 0.14 [95% CI -0.009, .30], p=.065).

Group comparisons – completing participants only

In-game assessments

Table S3 shows the mean performance on the in-game speechreading assessments (total, trained items and untrained items) from the 37 children

(Speechreading = 18; Maths = 19) who completed all of the training sessions. ANCOVAs were run on in-game assessment performance (all trials; trained model trials; untrained model trials) at the end of the intervention (post-intervention), with performance on the first in-game assessment (pre-intervention) as a covariate and group (intervention vs. control) as a fixed factor. Looking at all trials together, there was a moderate effect in favour of the speechreading training group ($F(1, 34)=3.08$, $p=.09$, $d=0.45$). Looking separately at the trained vs. untrained model trials, there was a large and significant effect of speechreading training on trained model trials ($F(1, 34)=4.70$, $p=.04$, $d=0.80$) but only a small, non-significant effect on the untrained model trials ($F(1, 34)=1.04$, $p=.32$, $d=0.17$).

Offline outcome measures

As already described, intervention compliance was variable. This is likely to have reduced the effectiveness of the intervention. A similar number of children completed each type of training: Speechreading $N = 18$; Maths $N = 19$. Therefore, for those variables where effects of the speechreading intervention were indicated in the intention to treat analyses involving all participants, we carried out additional exploratory analyses to examine whether effect sizes were larger in the subset of participants who completed the full intervention.

In all cases, larger effect sizes were seen for the group comparisons within the subset who completed the intervention compared to those observed when all participants were included. The means, *SD*s and 95% confidence intervals for each of these measures are shown in Table 3. After adjusting for performance at T1, there was a large effect of speechreading training on both outcomes of the ToCS Everyday Questions task at T3 (d_{Items}=1.29; d_{Words}=0.78;) and medium sized effects on both measures at T4 (d_{Items}=0.59; d_{Words}=.55;) as well as at T2 for the 'Items correct gist' measure (*d*=0.68). The effect of training on phonological awareness was moderate at T3 (*d*=0.34) and small at T2 (*d*=0.25) and T4 (*d*=0.24). There was no evidence of an effect of training on word reading at T2, T3 or T4 (*d*s=-0.01). There was a medium sized effect of training at T2 on overall vocabulary (*d*=0.46) and spoken vocabulary (*d*=0.36), but not at T3 or T4 (*d*s=-0.09, -0.13). The effect of training on AV speech production was small at T3 (*d*=0.27) and T4 (*d*=0.19).

Table 3. Comparison of participants who completed all training sessions – Intervention (N=18); Active Control (N=19).

	Intervention (*N* = 18)		Active control (*N* = 19)		Cohen's d^+	β (*p*) [95% CI]
	M	*SD*	*M*	*SD*		
ToCS Total Standard Score						
T1	94.11	9.46	95.89	12.89		
T2	92.83	10.37	93.95	10.88	0.06	-0.45 (.89) [-7.08, 6.19]
T3	93.83	10.55	96.16	12.40	-0.05	-1.41 (.68) [-8.18, 5.36]

ToCS Everyday Questions						
Words identified (max=62)						
T1	6.78	10.33	4.16	6.53		
T2	11.44	10.68	7.16	10.69	0.19	1.73 (.46) [-2.42, 6.05][1]
T3	17.39	12.23	8.05	12.27	0.78	6.80 (.07) [1.57, 13.40][1]
T4	20.44	11.86	13.05	15.24	0.55	4.80 (.23) [-1.47, 11.81][1]
Items correct gist (max=12)						
T1	0.50	1.15	0.37	0.83		
T2	1.28	1.53	0.47	1.02	0.68	0.72 (.08) [-0.08, 1.45][1]
T3	2.17	2.38	0.74	1.59	1.29	1.30 (.06) [-0.01, 2.65][1]
T4	2.72	2.42	2.00	2.29	0.59	0.57 (.41) [-0.92, 1.96][1]
Vocabulary (max = 74)						
Overall						
T1	54.89	5.73	56.16	6.69		
T2	65.83	5.57	64.21	4.43	0.46	2.21 (.13) [-0.86, 5.20][1]
T3	64.89	7.76	65.47	5.67	0.11	0.05 (.97) [-3.92, 3.59][1]
T4	63.94	6.39	65.79	4.09	-0.09	-1.34 (.40) [-4.55, 1.88]
Spoken						
T1	45.50	15.53	39.00	21.16		
T2	58.78	18.10	45.58	23.82	0.36	6.20 (.01) [2.07, 10.19][1]
T3	59.00	18.53	50.16	22.56	0.13	2.06 (.43) [-2.55, 6.88][1]
T4	59.61	16.66	53.58	19.78	-0.03	0.15 (.94) [-4.52, 4.56][1]
AV Speech Production (%) Updating[2]						
T1	68.62	30.69	53.06	29.86		
T2	74.56	26.68	55.16	32.72	0.13	4.66 (.27) [-2.75, 12.98][1]
T3	78.83	27.03	55.05	33.09	0.27	8.51 (.05) [1.47, 17.21][1]
T4	80.88	27.28	59.47	33.44	0.19	7.16 (.19) [-1.74, 17.91]
Phonological Awareness (max = 24)						
T1	10.78	4.14	10.53	4.68		
T2	13.83	5.59	12.47	5.17	0.25	1.12 (.32) [-1.12, 3.36]
T3	15.11	6.11	13.37	5.71	0.34	1.52 (.31) [-1.49, 4.53]
T4	15.78	5.71	14.47	5.23	0.24	1.10 (.41) [-1.51, 3.79][1]
Letter-sound Knowledge (max = 17)						
T1	12.06	4.39	11.05	5.79		
T2	13.83	4.18	12.84	5.90	-0.004	0.08 (.91) [-1.28, 1.52][1]
T3	14.17	4.81	12.58	6.15	0.11	0.60 (0.41) [-0.88, 2.00][1]
Word Reading (Z-Score composite)						
T1	0.55	3.33	0.12	2.32		
T2	0.76	2.94	0.36	2.52	-0.01	0.02 (.97) [-0.70, 0.73]
T3	0.86	2.86	0.46	2.49	-0.01	0.05 (.91) [-0.82, 0.92]
T4	0.80	2.67	0.41	2.28	-0.01	0.11 (.85) [-0.94, 1.13][1]
Number Skills (Z-Score composite)						
T1	0.19	2.28	0.22	1.98		
T2	0.45	2.09	0.27	1.98	0.10	0.21 (.59) [-0.56, 0.92]
T3	0.35	2.00	0.43	1.96	-0.02	-0.05 (.91) [-0.87, 0.78]

[+]Cohen's *d*: Difference in progress between groups divided by pooled initial *SD*
[1]ANCOVA models run with bootstrapped SEs, bootstrapped bias-corrected and accelerated 95% CIs are reported
[2]N = 16 Speechreading, N = 17 Maths

Discussion

We examined the efficacy of a 12 week computerised speechreading training intervention for deaf children using a randomised controlled trial. Our first hypothesis was that we would see gains in speechreading skills following the speechreading intervention. There was no evidence of effects of the intervention on our pre-specified primary outcome variable, standard score on the Test of Child Speechreading, at any time-point post-intervention. However, the speechreading intervention group did show gains relative to the control group on the Everyday Questions speechreading test (a pre-specified secondary outcome variable), which involved untrained talkers and untrained items. This effect was large, and significant, in the intention to treat analyses including all participants 3 months after the end of training. Moderate gains were still evident 11 months after training, though these were no longer significant. At both time-points these effects were larger in exploratory analyses which included only those participants who completed all the training sessions.

That the evidence of a gain in speechreading skills was largest 3 months after training suggests that an interim period post-intervention may have been necessary for the full benefits of the intervention to be realised. Consistent with this, results on the in-game speechreading assessments showed that, by the end of the intervention, children who completed the speechreading training showed significant advantages relative to the children who completed the maths training when speechreading trained talkers but not untrained talkers. This suggests that transfer effects to speechreading of unfamiliar people may take time, and experience of using speechreading in the real world, to manifest.

To our knowledge this is the first RCT to evaluate a speechreading intervention with young deaf children. The finding of significant effects of training on a speechreading measure that involved untrained models and untrained stimuli (Everyday Questions test) in our intention to treat analyses suggests transfer of the training effects beyond the items and models that were included in the intervention and provides support for the efficacy of this computerised speechreading training programme in boosting speechreading skills in young deaf children.

The finding of a significant advantage to the speechreading intervention group on the Everyday Questions speechreading measure, but no effects of the intervention on the core Test of Child Speechreading may be explained by differences in response format between the two measures. The Test of Child Speechreading involved a forced choice, closed set response format in which children could guess the answer. By contrast, the Everyday Questions measure scored a free response, and as such may offer a more valid measure. However, the Everyday Questions task was difficult for many of the children which may have limited its sensitivity to detect changes in speechreading skill for the lower performers. Therefore, future studies of speechreading with children of this age may benefit from using a single word, free response speechreading task.

The data suggest that it is possible to train speechreading skills in young deaf children using a computerised online training programme. Further studies are needed to establish which deaf children will benefit most from this programme. However, the training programme is likely to be of interest to any Teachers of the Deaf, Speech and Language Therapists, or parents of deaf children who are looking for tools to improve a deaf child's speechreading skills.

The second hypothesis we sought to address was that gains in speechreading would lead to subsequent gains in phonological and reading skills. One measure of phonological skill was a measure of speech production which scored both auditory and visual components of the child's speech during picture naming of items which were included in the speechreading training. Children in the speechreading group who had poorer AV speech output at the start of the study, showed evidence of a significant benefit of speechreading training to their AV speech production, when compared to the control group. There was also evidence of a moderate effect of speechreading training on phonological awareness three months after the end of training, though this did not reach significance.

In summary our data suggest that short-term speechreading training was able to bring about longer-term gains in speechreading skills in young deaf children and there was some evidence of beneficial effects on phonological skills. Although these results are encouraging, especially in relation to the gains observed in speechreading, the gains in phonological skills were all small to moderate and not statistically significant in all cases. There was also no consistent evidence of an effect of the speechreading training on word reading. Although children improved in their reading proficiency over the course of the study their reading performance, relative to others in the sample, was highly stable over time. This indicates little influence of the speechreading training on individual trajectories of reading development.

There are a number of potential reasons why we did not see larger downstream consequences of the speechreading training. First, the complete speechreading programme only provided 8 hrs of speechreading training over the space of three months. This is in the context of the additional input that the children would have been receiving to foster their reading development during this time both at school and at home. Thus, the speechreading gains brought about by the speechreading intervention offered in this programme may not have been of sufficient magnitude to bring about detectable gains in reading on their own. It may be that speechreading training is most effective when fully embedded as part of a broader literacy programme. Relatedly, for this cohort of children, the majority of whom have increased auditory access to the phonology of spoken language compared to previous generations of deaf children, an intervention that capitalised on both available auditory and visual information about phonology may have been more effective in helping to develop their phonological representations. Aspects of the study design may also have limited its capacity to demonstrate benefits of the intervention. We selected children for this study based on age, rather than language level. Some children were already competent speechreaders and readers. It is likely that children with the poorest speechreading and reading skills are those who would benefit most from the training; though a minimum level of spoken language knowledge is also likely to be a pre-requisite for benefit. The study may also have been limited in its power to detect effects of the intervention due to the relatively small sample size, compounded by issues around adherence to the intervention. The issue of small sample sizes in studies with special populations is a common limitation; more cross-centre collaborative studies to increase participant numbers would be a valuable way to address this. Adherence to the intervention is also an issue that future studies should aim to address. Although conducted in schools, less than two thirds of children in each group completed the planned number of training sessions. Frequent issues encountered included consistency of support to help the child log on to the programme and technical issues with school IT systems.

Despite these issues, the study has confirmed the feasibility, for the first time, of conducting an RCT to examine the efficacy of literacy interventions for deaf children. In their review of the literature on strategies for teaching deaf children grapheme-phoneme correspondences, Tucci, Trussell and Easterbrooks (2014) highlighted the dearth of intervention studies in this area and argued that "the evidence base for literacy interventions in the field of deaf education is still in its infancy". Deaf children deserve the same high-quality evidence base to inform their literacy instruction as hearing children and increasing the size and quality of that evidence base should be a priority.

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Key points

- Learning to read is a difficult task for most deaf children. Previous studies have demonstrated that speechreading (lipreading) is a good predictor of reading in young deaf children.
- We developed and assessed the effects of a computerised speechreading training programme on speechreading, phonological and reading skills in young deaf children.
- We found significantly greater gains in speechreading performance in deaf children who carried out speechreading training compared those who completed maths training.
- There was also some evidence of beneficial effects of speechreading training on phonological skills, but not reading.

- These findings demonstrate that speechreading skill is trainable in young deaf children, however to be effective at supporting early reading this may need to be embedded in a broader literacy programme.

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Supplementary Methods

Speechreading training

Stimuli

103 words were included in the training dataset. All words were concrete nouns and were chosen because of their early age of acquisition. Art work was created for the games to represent the items (for examples, see [*Figure S1*]). These images were refined following tests of naming agreement with hearing 4-5yr old children. Four different talkers (three adults, one child; 2M,2F) were filmed saying each of the English spoken labels for the items aloud. Although the stimuli were recorded audio-visually, participants only ever saw visual-only videos of the spoken words. Children saw all four talkers saying all words throughout the course of the training to encourage them to learn to extract the commonalities between visual speech patterns of different talkers.

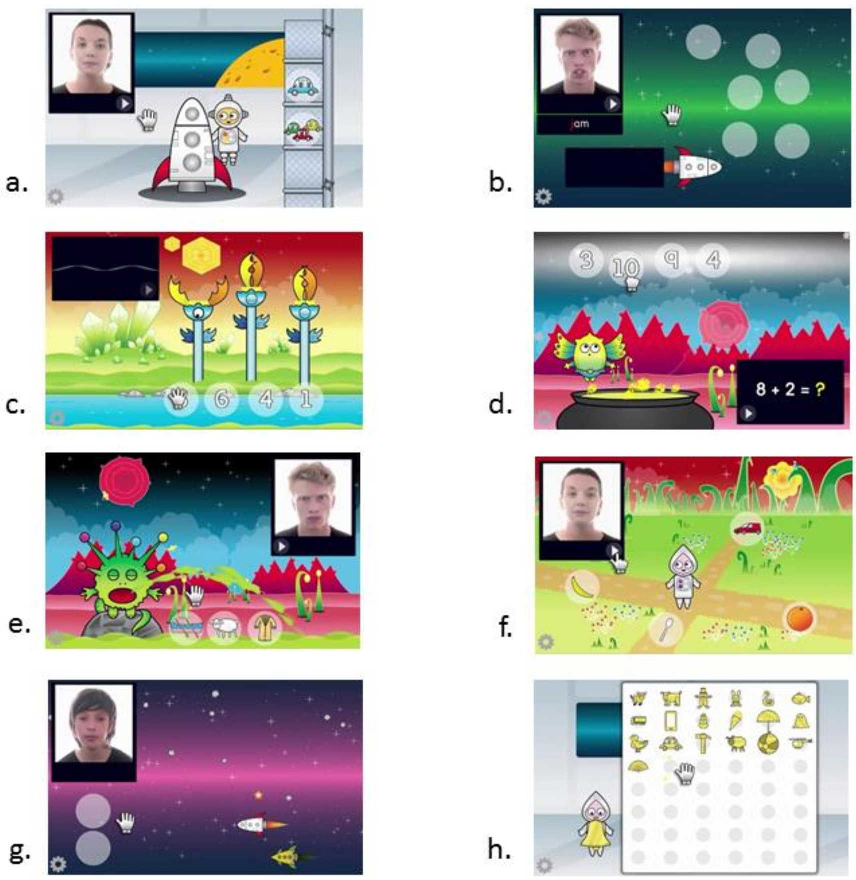
Game design

At the beginning of each 10 minute training session the children completed a brief task that was designed to help them understand the relevance of good speechreading conditions in the real world. In these tasks the children had to get the models that they would see in the games ready so they could speechread them. For example, in one task they had to press a button that gradually turned up the light on one of the speechreading models until they could see their face well enough to speechread. In another, they had to press a button to make the model turn around until they were facing them.

The speechreading intervention comprised algorithm-based speechreading and reading training, set within the context of seven space-themed computer games (see Figure S1 for examples). The training was designed to run across 48 10 minute sessions. The first 16 sessions contained trials that involved visual speech videos and pictures only. These focused on introducing the vocabulary used in the intervention (103 words) and on mapping speechread words to a corresponding image. In these trials, children saw a silent video of a model saying the target word (e.g. 'rabbit') and then saw a speech bubble overlaid on the video with the corresponding target image in it (i.e. they were given an explicit pairing of the visual speech token and a picture that the token referred to). They could then choose the correct target image from two response options. Immediately following this they would do a paired trial in which they would see the target image from the previous trial, and had to choose the corresponding video with the target visual speech from a choice of two video response options (e.g. 'rabbit' and 'elephant'). For all trials, participants were free to articulate the perceived words if they chose to do so.

For the core speechreading trials, children saw a video of a model saying one of the 103 target words and then had to choose the corresponding picture from a choice of the target and three distractors. An algorithm was developed that enabled the difficulty level of these trials to be systematically varied in an adaptive way based on the child's performance. The adaptive algorithm was driven by varying the visual similarity between the target and the distractors based on the visual similarity of their constituent phonemes. To derive this visual similarity information, we collected data from British English-speaking hearing adults using an established paradigm (Auer & Bernstein, 1997) to determine the confusability of individual phonemes presented in the visual-only modality. Participant visual phonemic identification data produced confusion matrices (separately for vowel and consonant phonemes). Multidimensional scaling solutions were then applied to the confusion matrices to estimate visual phonetic similarity. To provide information for the speechreading algorithm on how visually similar any two words from the pool of 103 were, Similarity Choice Model similarity coefficients for each possible pair of phonemes were calculated. These allowed an estimate of the visual similarity between each of the 103 stimulus words and every other word based on the similarity of the constituent phonemes.

Figure S1: Screenshots from each of the seven computer games (a. to g.) that were used in the intervention and control conditions of the trial and from one of the reward scenes (h.). Example content from both the speechreading and number and maths interventions is shown in the seven games.



Creating the adaptive algorithm in this way meant that children would begin with targets and distractors that were highly visually distinct and would advance through to targets and distractors that were progressively more similar when they achieved criterion levels of success on easier trials. An example of progressively more difficult contrasts is: bee-fish > bee-boot > bee-bees> bee-pea. An example of an easy trial would be to match the spoken target 'mat' to images of 'mat, elephant, spoon, car', in which the overlap in visual speech between target and distractor pictures is very low. An example of a difficult trial would be to match 'mat' to the target picture, selecting from 'mat, map, hat, pan' in which the visual speech overlap is high.

In addition to trials operating at the single word level, children also completed trials which a) showed videos of two word utterances (e.g. 'red hat'; 'blue door') and b) showed videos of the two word utterances within a carrier sentence and hence required the child to disembed the key information from the surrounding sentence (e.g. 'find the red hat this time'). In both cases, these trials still involved video to picture matching.

Sessions 17 through 48 continued the speechreading training trials introduced in the first 16 sessions but additionally included trials that contained orthographic stimuli and that focused on training mappings between visual speech patterns and letters and words. These trials were designed to use visual speech to target the skills of grapheme-phoneme matching (e.g. seeing a video of a phoneme and choosing the corresponding letter or digraph), blending and segmenting (e.g. seeing a video of a word broken down into its constituent phonemes and choosing a picture that corresponded to the blended whole word), and spelling (e.g. seeing a video of a whole word and picking letters to spell that word).

The reading trials were rendered adaptive in two ways. First, the level of support was varied such that children moved through a systematic series of levels of difficulty on the same stimulus. For example, on easier blending trials the visual speech stimuli were accompanied by simultaneous corresponding orthographic stimuli. On more difficult trials the visual speech stimuli were presented with orthography and children had to derive the orthographic correspondence without support. On easier spelling trials the words were broken down into their constituent phonemes and then blended to make the whole word. On more difficult trials the whole word was presented and the children had to segment the word themselves to complete the spelling task.

A second way in which the reading algorithm operated adaptively was by varying the complexity and regularity of the orthographic to phonological mapping of the words used. The words in the intervention were divided into six pools. Pool 1 contained words that were CVC in structure and contained regular orthography-phonology mappings involving a single letter to a single sound (e.g. 'pig', 'tap', 'zip'). Pool 2 contained words that contained regular orthography-phonology mappings and included consonant digraphs in addition to single letter to single sound mappings (e.g. 'chip', 'fish', 'king'). Pool 3 contained words that contained regular orthography-phonology mappings and included vowel digraphs in addition to single letter to single sound mappings (e.g. 'coat', 'moon', 'tree'). Pool 4 contained words that had one or more complex or irregular orthography-phonology mappings (e.g. ‘ball’, ‘knee’, ‘wheel’). Pool 5 contained words that contained split digraphs (e.g. ‘bone’, ‘cake’, ‘kite’). Finally, pool 6 contained words that had complex mappings, were multisyllabic or did not fit in one of the previous pools (e.g. ‘elephant’, ‘scissors’, ‘trousers’). Reaching a pre-specified criterion level of success on each pool of words enabled the children to progress the subsequent pool.

Active Control Condition (Maths training)

The children in the control group played the same set of seven space-themed computer games as the children in the speechreading group, however the content of the games was number and maths trials not speechreading. Therefore, children in the two groups experienced the same visual environment and rewards, with the only difference being the skills being trained in the games. The maths content was driven by adaptive algorithms that presented early number skills, counting, and arithmetic trials that responded to the child’s performance level.

Difficulty level was varied both by the numbers used (e.g. 1-10 vs. 10-20) and the operations required on those numbers. For example, moving from mapping objects to objects to mapping objects to digits; moving from completing sequences where numbers count up in 1 to sequences where numbers count up in 5; moving from completing additions where the sum remains on the screen to completing additions where the sum disappears and has to be retained and operated on in working memory.

Assessments

In-game assessments

There were seven In-Game Assessments in total, with the first assessment completed prior to the first session and final assessment at the end of the 48 training sessions. Therefore, only those who completed all of the training sessions, completed all of the In-Game Assessments. In each assessment trial the children viewed a video of a model saying one of the trained words and had to choose the corresponding picture from a choice of four. There were 30 trials in total, 15 of which used videos of the models from the speechreading intervention (trained) and 15 parallel trials with the same target word and response options but which used videos of a model who was not included in the speechreading intervention (untrained). These in-game assessments were completed independently by the children during the training sessions and not administered by the researchers.

Assessments at pre-test (T1), post-test (T2), after intervention follow up at 3 months (T3) and 11 months (T4)

Pre-specified *primary outcome measure: Test of Child Speechreading (ToCS) – core test*

The ToCS core test starts with a familiarisation task in which children see a silent video of the two models who produce the test stimuli saying the days of the week. Each of the three subtests follows a similar format, beginning with practice trials in which explicit feedback is given, followed by test trials in which no feedback is given. The children watch a silent video of a model saying a word, sentence or short story and then must choose a picture which corresponds to their answer from a choice of four. For the words and sentences subtests, the picture chosen must correspond to what the model said. For the short stories part of the assessment, the tester asks the child two questions about each story and they must choose a picture that answers the question asked. There are 15 trials in the words and sentences subtests and 10 in the short stories subtest, giving each child a total raw score out of a possible 40.

The following pre-specified *secondary outcome measures* were also collected.

Speechreading: Test of Child Speechreading (ToCS) – Everyday Questions test

Children were required to watch silent videos (n=12) of two talkers asking questions they might encounter in everyday life (e.g. where do you live?) and tell the experimenter what they thought the question was. Children could answer using their preferred communication mode. Children received two scores on this measure, one reflecting the number of questions they correctly reproduced the gist of (ToCS Everyday Questions Items Correct Gist), and one reflecting the total number of individual words that the child got correct across all 12 questions out of a possible 62 (ToCS Everyday Questions Words Identified). For example, if the question was 'how old are you?' and the child's response was 'how are you?', they would receive 0 on that item for the Items Correct Gist score but 3 for the Words Identified score. If the question was 'what did you eat for breakfast?' and the child's response was 'what did you have for breakfast?', they would receive 1 on that item for the Items Correct Gist score and five (out of a possible six) for the Words Identified score. The responses were transcribed online during the testing session, checked and scored offline from the video by the tester, and then checked from the video by a second blinded scorer.

Vocabulary

A naming task, using the pictures from the training, was used to assess participants' knowledge of the vocabulary used in the speechreading training. Their first response was taken for each trial. If they named it in sign, they were asked if they knew the English word. Each participant was given a score for the number of correct items produced in spoken English (Spoken Vocabulary; total = 74) and a score for the number of correct items produced either in spoken English or BSL, thus providing a measure of overall vocabulary, regardless of modality (Overall Vocabulary; total = 74).

Audio-visual speech production (AV speech production)

Participants were filmed completing the picture naming task described above. For the purposes of obtaining a speech production score, if the child named the picture incorrectly or could not name it at all on their first attempt, the experimenter provided them with the correct label and asked them to repeat it.

The 30 words selected for this measure were chosen to maximise the range of phonemes in syllable-initial and syllable-final positions, and to provide a range of word lengths and syllable structures, including consonant clusters. To calculate a score that reflected changes in the quality of phonological representations of the same words over time for each child, items that were named incorrectly or not attempted at any of the time points were excluded from the analysis. Attempts that were phonologically unrelated to the target word were also excluded, to avoid random vocalisations. Thus, each child received a total possible score based on the words that they attempted at all time-points. This was used to obtain their overall score at each time point as a percentage of the possible score.

A narrow transcription was made for each word, based on the International Phonetic Alphabet (IPA). Each consonant was then scored according to the following scoring system: Correct within the boundaries of the target phoneme or an acceptable allophone, including accent variations (4 points); Place correct plus either voice or manner correct (3 points); Place correct but voice and manner are incorrect or for all target consonants further back than dental, place not correct but within the wider category (i.e. coronals, velars, glottals) or silent articulations in the correct place or place in the wider category or clicks in the correct place or place in the wider category (2 points); place incorrect or (for target consonants further back than dental), not within the wider category (1 point); omission (0 points). The maximum score for each consonant was 4, and each word had a maximum score based on the number of consonants. The maximum total for all 30 words was 284. To verify the reliability of transcriptions, a second marker transcribed and scored a subset of 10% (6 children/540 words/1278 consonants) of the data.

Agreement between the two scorers was good (Cohen's Kappa = 0.71, SE = 0.02).

Phonological awareness

In the onset trials (n=12), children viewed a target picture (e.g. house) and had to choose the item from a choice of three (e.g. hand; cow; jam) that started with the same sound. One of the incorrect distractors overlapped with the target in terms of vowel (near distractor; e.g. cow). The rime trials (n=12) followed the same format. In this case, the correct response (e.g. peg) shared the rime with the target (e.g. leg). The near distractor shared the vowel with the target (e.g. bell).

Letter-sound knowledge

The letter-sound productions were scored online during the testing session, checked offline from the video by the tester, and subsequently checked offline from the video by a second blinded scorer. This assessment was not carried out at T4.

Word reading

Three measures were used to assess the children's word reading ability. The first two were taken from the YARC (Snowling et al., 2009) and assessed single word reading of untrained stimuli. The early word recognition test (EWRT) is designed for 4-7 year olds and assesses children's ability to read 30 early acquired words. The single word reading test (SWRT) was also administered to avoid any ceiling effects as it was designed for 5-11 year olds and hence contained more challenging words (n=60). Children who used BSL as their preferred communication mode labelled the word in sign rather than reading it aloud in English. These reading measures were scored online during the testing session, checked offline from the video by the tester, and subsequently checked offline from the video by a second blinded scorer.

The third reading measure was a novel test that assessed single word reading for stimuli included in the speechreading training (n=24 trials). Children saw a word in the middle of the screen and had to point to the corresponding picture from a choice of four, therefore no speech production was required. A reading composite score was created by summing each child's z scores on the three word reading measures.

Number skills

Three measures of number skills were administered. 1) The Early Number Concepts section of the BAS-III (Elliot & Smith, 2011) provided a measure of children's understanding of concepts related to number (e.g. 'more than', ‘less than’) and early number skills (e.g. counting, adding, subtracting). 2) A standardised measure of addition and subtraction fluency taken from the Test of Basic Arithmetic and Numeracy Skills (Hulme, Brigstocke, & Moll, 2016). 3) Children were asked to count to 30, with the highest number they could reach being their score on this task. A Number Skills composite score was created by summing each child’s z scores on the three measures of number skills. At T4 only the measure of addition and subtraction fluency was administered.

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Table S2: Intention to treat analyses: Means and SDs, for all participants on the outcome measures at Baseline (T1) and the Follow-up, 11 months after training (T4). Also presented are Cohen's *d* effect sizes and results of the ANCOVAs comparing the two groups on each outcome at T4 while adjusting for their baseline performance.

	Intervention (N = 32)		Active control (N = 30)		+Cohen's *d*	β (*p*) [95% CI]
	M	SD	M	SD		
ToCS Single Words Subtest (max = 15)						
T1	7.28	2.49	7.60	2.61		
T4	8.75	2.84	8.70	2.87	0.14	0.25 (.69) [-0.98, 1.47]
ToCS Everyday Questions						
Words identified (max = 62)						
T1	5.50	9.27	5.90	11.41		
T4	18.13	12.38	15.37	17.96	0.31	3.18 (.29) [-2.93, 8.81][1]
Items correct gist (max = 12)						
T1	0.41	0.95	0.80	2.04		
T4	2.41	2.27	2.47	3.10	0.22	0.41 (.43) [-0.60, 1.42][1]
Vocabulary (max = 74)						
Overall						
T1	54.50	5.78	53.57	9.47		
T4	63.34	6.11	63.33	7.51	-0.12	-0.52 (.70) [-3.19, 2.16]
Spoken						
T1	41.78	17.15	37.60	21.50		
T4	55.47	18.55	52.60	19.21	-0.08	-0.88 (.65) [-4.70, 2.95]
AV Speech Production (%) Updating[2]						
T1	65.67	31.97	59.74	30.48		
T4	74.57	32.95	68.10	31.91	0.02	0.92 (.81) [-6.15, 8.74][1]
Phonological Awareness (max = 24)						
T1	10.28	4.23	10.77	5.19		
T4	14.59	5.36	14.30	5.57	0.17	0.70 (.48) [-1.27, 2.66]
Word reading (Z-score composite)						
T1	0.01	2.89	0.05	2.95		
T4	0.29	2.58	-0.31	3.05	0.22	0.64 (.16) [-0.25, 1.53]
Arithmetic fluency (max = 60)						
T1	4.25	4.71	4.77	6.07		
T4	13.19	10.17	14.00	11.95	-0.05	0.12 (.92) [-2.63, 2.95][1]

+Cohen's *d*: Difference in progress between groups divided by pooled initial SD

[1]ANCOVA models run with bootstrapped SEs, bootstrapped bias-corrected and accelerated 95% CIs are reported

[2]N = 30 in each group

Table S3: Means and SDs for performance on the In-Game Assessments (N=7) for the children who participated in all of the training sessions. Data for overall performance and on the trained and untrained models separately are provided.

		Intervention (*N* = 18)		Active control (*N* = 19)	
		M	SD	M	SD
IGA 1	Total	12.83	3.87	11.58	3.79
	Trained	6.83	1.76	6.47	2.61
	Untrained	6.00	2.89	5.11	1.59
IGA 2	Total	13.83	5.20	11.63	4.22
	Trained	7.17	3.03	6.00	3.04
	Untrained	6.67	2.72	5.63	1.86
IGA 3	Total	14.11	3.76	13.16	4.41
	Trained	7.56	2.28	6.79	2.78
	Untrained	6.56	1.89	6.37	2.24
IGA 4	Total	14.33	5.03	13.26	4.62
	Trained	7.44	2.83	6.95	2.55
	Untrained	6.89	2.49	6.32	2.81
IGA 5	Total	14.83	4.31	12.68	5.02
	Trained	7.67	2.70	6.47	2.89
	Untrained	7.17	2.33	6.21	2.44
IGA 6	Total	14.94	5.62	11.95	3.95
	Trained	8.11	3.10	6.00	2.43
	Untrained	6.83	2.98	5.95	2.50
IGA 7	Total	14.72	4.69	11.74	4.19
	Trained	7.39	2.68	5.68	2.16
	Untrained	7.33	2.40	6.05	2.57

IGA = In-Game Assessment; Total Max=30; Trained/ Untrained Max =15

Appendix B

Table A. Stimuli lists of trained and untrained items for Chapter 8

Untrained set	Trained set
dog	cow
hat	ball
cake	bath
fish	boot
fork	hand
shoe	leaf
star	road
tree	rope
cloud	shop
clown	sock
house	chair
light	clock
snake	dress
toast	ghost
banana	knife
carrot	spoon
rabbit	monkey
spider	glasses
snowman	icecream
aeroplane	telephone

NB. Words were presented in different orders for the speechreading, reading, and phonological awareness tasks based on the requirements for the task. Words are presented here in order of word length.

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