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Title: Literacy and phonological skills in oral deaf children and hearing children with a history of dyslexia

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Literacy and phonological skills in oral deaf children and hearing children with a history of dyslexia

Abstract

Oral deaf children and hearing children with dyslexia both experience literacy challenges, although their reasons differ. This paper explores the problems underlying poor literacy in each group and draws implications for reading interventions.

Data were collected using standardised literacy and phonological measures from 69 severe-profoundly prelingually-deaf children aged 10-11 years, all communicating with spoken language, and compared with equivalent data from 20 hearing children with a history of dyslexia matched on reading ability. Children were given a large battery of tasks assessing word and nonword reading, spelling, vocabulary and reading-related skills including letter-sound knowledge, phonological awareness, rapid automated naming and verbal short-term memory. Striking similarities were observed for word reading, nonword reading and spelling across groups, and associations between the measures and reading-related skills were similar. However, differences between the two groups emerged in the strength of association between literacy and vocabulary. Regression analyses confirmed vocabulary as a key predictor of literacy in the oral deaf group.

These results suggest that not only children with a history of dyslexia but also oral deaf children who struggle with reading should receive specialist literacy support. Reading interventions for oral deaf children should target phonological and language skills within a fully integrated approach.

Introduction

Dyslexia and accompanying deficits have been widely researched (Bishop & Snowling, 2004; Lyon, Shaywitz & Shaywitz, 2003; Peterson & Pennington, 2015; Ramus, 2003;

Saksida et al., 2016; Snowling, 2000, 2008; Vellutino, Fletcher, Snowling & Scanlon, 2004). With increasing awareness of dyslexia, a diagnosis of dyslexia in the UK can lead to recognition of children's needs and the provision of specialist support and intervention (Duff & Clarke, 2011). However, the same cannot be said for deaf¹ children, many of whom are widely acknowledged to have severe reading and language delays (Conrad, 1979; Harris, Terlektsi & Kyle, 2017; Herman, Kyle & Roy, 2016; Kyle & Harris, 2006; 2011; Wauters, Van Bon & Tellings, 2006). Drawing on theoretical models of equifinality, whereby 'the same end state may be reached from a variety of initial conditions and through different processes' (Cicchetti & Rogosch, 1996, p. 597), versus multifinality (ibid.), where the same aetiology may lead to different outcomes, this paper compares the reading profiles of these two groups of struggling readers: hearing children with a history of dyslexia and oral deaf children who communicate primarily using spoken language. We seek a better understanding of the impact of their respective developmental pathways by exploring similarities and differences in the reading profiles of these groups, with implications for interventions for each, in particular for struggling oral deaf readers, for whom currently no specific reading interventions exist.

Reading difficulties in hearing children

Several different profiles have been proposed for hearing children with reading difficulties, exemplified using the Simple View of Reading (SVR, Gough & Tunmer, 1986). In this model, decoding skills entail matching speech sounds to written letters and rely on an established speech sound (phonological) system and use of letter-sound correspondence rules, whereas language comprehension skills, comprising lexical and syntactic knowledge, are needed to understand decoded words, become literate and read with meaning. Following the SVR, dyslexic children typically present with good language but weak decoding skills,

¹ We use the term 'deaf' here to refer to individuals with a prelingual severe-profound degree of hearing loss, i.e., one that is present at or shortly after birth.

whereas poor comprehenders demonstrate age-appropriate decoding but weak language comprehension skills (Bishop, McDonald, Bird & Hayiou-Thomas, 2009; Bishop & Snowling, 2004). Gough and Tunmer (1986) identified a further group of struggling readers, ‘garden variety’ poor readers (p. 8-9), who exhibit both low decoding and weak language comprehension skills that they argued comprised the majority of poor readers. While reading and language difficulties may co-occur and there may be an overlap in behavioural manifestations, the underlying causes of language and reading impairments can be quite distinct (Bishop & Snowling, 2004).

Our focus in this paper is on the decoding aspect of learning to read, which presents a particular challenge for children with dyslexia. In dyslexia, poor decoding is related to difficulties in verbal short-term memory and verbal processing skills, and poor recognition and manipulation of sound patterns (phonological awareness) (Rose, 2009; Snowling, 2008; Vellutino et al., 1996). Together, these phonological deficits have been seen as the core deficits in dyslexia (Hulme & Snowling, 2009; Petersen & Pennington, 2015; Ramus, 2003; Saksida et al., 2016; Snowling, 2000). Although a consensus definition of dyslexia remains contentious, with some using the term to describe any child who struggles with reading (see Elliott & Grigorenko, 2014, for a fuller discussion), it is the phonological model of dyslexia that we adopt in the current study. Using this model, difficulties with accurate and fluent word recognition and spelling experienced by children with dyslexia are seen to arise as a consequence of phonological deficits, despite normal intelligence and adequate instruction (Lyon et al., 2003; Tunmer & Greaney, 2010).

However, as Gough and Tunmer (1986) state, this begs the question of why their phonological skills are weak. Most argue for a biological link of some kind. It is generally agreed that phonological deficits are constitutional problems and genetically based, particularly in cases where they are severe (Bishop & Snowling, 2004), although

environmental factors such as socioeconomic status are also significant (Fuller-Thomson & Hooper, 2014; Snowling, 2008; Snowling, Muter, & Carroll, 2007).

Prelingual deafness and reading

Reading difficulties in deaf children have also stimulated research interest and afford an alternative developmental pathway for weak decoding skills. Prelingual deafness, (i.e., a significant permanent hearing loss that is present from birth) imposes a barrier to accessing speech sounds and limits exposure to spoken language, upon which written language is based, with repercussions for language development. Although most deaf children (67%) communicate orally, i.e., use spoken language, a minority (7%) use a sign language such as British Sign Language (BSL), and a significant proportion (26%) use a combination of modes, typically the use of key signs alongside spoken language, known in the UK and USA as Sign Supported English or SSE (Consortium for Research in Deaf Education, 2017). Although there are no barriers to deaf children accessing a visuo-gestural language such as sign, sign language acquisition is impacted by poor language exposure as most deaf children have hearing parents who, in comparison to deaf parents, typically come to sign language late and struggle to achieve high levels of fluency (Woolfe, Herman, Roy, & Woll, 2010). As a consequence, many signing deaf children frequently experience significant language delays (ibid).

Prelingually deaf children's reading often develops at a slower rate than their typically developing hearing peers (Allen, 1986; Harris, Terlektsi & Kyle, 2017; Kyle & Harris, 2010), culminating in the average deaf school leaver (i.e., upon leaving secondary school at 16 years) reported as having a reading age approximately equivalent to that of a nine year-old hearing child (Conrad, 1979; Wauters et al., 2006). Like dyslexic individuals, deaf children are frequently reported to have weaker phonological skills in comparison with typically developing hearing peers, both in terms of phonological awareness and phonological coding

(Cupples, Ching, Crowe, Day & Seeto, 2013; Johnson & Goswami, 2010; Kyle & Harris, 2006; Nitttrouer, Sansom, Low, Rice & Caldwell-Tarr, 2014). Despite overall lower levels of ability, there is increasing evidence that better phonological skills are associated with higher levels of reading in deaf children, in the same way they are in hearing children (Campbell & Wright, 1990; Colin, Magnan, Ecalle & Leybaert, 2007; Cupples et al., 2013; Dyer, MacSweeney, Szczerbinski, Green & Campbell, 2003; Easterbrooks, Lederberg, Miller, Bergeron & Connor, 2008; Harris & Beech, 1998; Lederberg, Schick & Spencer, 2013; Mayer, 2007), particularly in oral deaf children and children with cochlear implants (Dillon et al., 2012; Johnson & Goswami, 2010; Spencer & Tomblin, 2009). For deaf children who use sign language, there are contradictory views over whether phonological skills are significant (Hirshorn, Dye, Hauser, Supalla & Bavelier, 2015; Mayberry, del Giudice & Lieberman, 2011), with some studies suggesting that signing deaf children may develop phonological skills through the process of learning to read (Kyle & Harris, 2010; Goldin-Meadow & Mayberry, 2001), mirroring an earlier debate on the directionality of effects in the hearing population (Castles & Coltheart, 2014; Hulme, Snowling, Caravolas, & Carroll, 2005), and others arguing that phonological skills are not relevant (e.g. Belanger, Baum & Mayberry, 2012; Clark et al, 2016), with implications for the applicability of the SVR to deaf children.

A further issue is that the development of phonological representations in deaf children proceeds differently to that of hearing children. Whereas hearing children's representations arise primarily from listening to spoken language, deaf children additionally use visual cues from speech-reading (Kyle & Harris, 2010; 2011; Kyle, Campbell & MacSweeney, 2016). Cued Speech², Visual Phonics³, finger-spelling⁴ and alphabetic script may also underpin phonological development and lead to better levels of reading ability (e.g., Haptonstall-Nykaza & Schick, 2007; Leybaert, 2005; McQuarrie and Parilla, 2009; Narr, 2008; Trezek,

² Cued Speech is a visual version of English based on a system of handshapes and positions used while speaking to disambiguate lip-patterns (www.cuedspeech.co.uk n.d.)

³ Visual Phonics is a series of hand cues and written symbols used to represent the individual sounds of English used only in phonics teaching (International Communication Learning Institute, 1996).

⁴ Finger-spelling is the manual alphabet used in sign languages.

Wang, Woods, Gampp & Paul, 2007). Phonic based interventions using Cued Speech and Visual Phonics have been used with some success to improve reading levels of struggling deaf readers (Miller, Lederberg & Easterbrooks, 2013; Trezek & Malmgren, 2005; Trezek & Wang, 2006) and provide further evidence that, for deaf and hearing children alike, phonological knowledge of the written language is important if they are to become competent readers (Mayer, 2007).

Although many deaf children find reading to be a difficult task, multifinality in reading outcome is evident, since some do become proficient readers. Higher levels of reading achievement have been reported in studies with selective populations of deaf children, such as those educated orally (Gravenstede & Roy, 2009), children with cochlear implants (e.g. Archbold et al., 2008; Dillon, de Jong & Pisoni, 2012; Marschark, Rhoten & Fabich, 2007; and see review by Mayer & Trezek, 2017), those with deaf parents (Strong & Prinz, 2000; Vernon & Koh, 1970) and those with early exposure to good models of American Sign Language (ASL) (Mayberry 2007, 2010; Petitto, 2009; Scott & Hoffmeister, 2016). Furthermore, studies with deaf adults have reported higher levels of literacy in those who were exposed to ASL from an early age (Chamberlain & Mayberry, 2008; Mayberry, 2007; Stone et al., 2015). However, many of these studies look at adults and older secondary-aged deaf children who are bilingual in ASL and spoken English, with reading comprehension as the main outcome measures, where as noted above, language skills as opposed to decoding skills are particularly significant. Indeed, it is unclear from the majority of studies whether the strong association between signing skills and reading actually results in age appropriate reading levels. What is clear from extant research is that while there is no consensus for the role of phonology in deaf children's reading, there is little debate over the crucial role of underlying language skills in proficient deaf reading, regardless of the whether these underlying language skills are in sign or spoken language (see Harris et al., 2017; Kyle, 2018). Therefore, although findings from research with signing deaf children can inform our

understanding of reading in oral deaf children, especially with respect to the role of language, our main focus in the current study is primarily on decoding skills in a selected sample of orally educated deaf children.

Comparing deaf children and hearing dyslexic children

Although both dyslexic and deaf children have difficulty with phonological skills, the extent to which these skills underlie literacy performance in each group is not clear. Whilst the heterogeneous nature of dyslexia is generally recognised (Petersen & Pennington, 2015), particularly in unselected samples (Carroll, Solity & Shapiro, 2016), the key role of phonological deficits in dyslexic children is well established (Bishop & Snowling, 2004; Petersen & Pennington, 2015). However, the degree to which such deficits underpin weak literacy in deaf children, and more generally the role of phonological skills, is less well understood.

Only a few studies to date have compared deaf and hearing dyslexic readers directly and three of these have focused exclusively on the contribution of underlying deficits in the magnocellular visual system to reading difficulties in deaf adults (Samar & Parasnis, 2005; 2007; Samar, Parasnis & Berent, 2002). Samar and colleagues (2002; 2005) suggested that all struggling deaf readers could be thought of as dyslexic on the basis of these visual deficits and thus dyslexia may be the largest secondary disability in the deaf population. However, the role of these and other visual deficits (e.g., visual attention span, visual tracking, etc.) in dyslexia among the hearing population is controversial; although they frequently co-occur, whether or not they are part of the underlying cause of dyslexia remains unclear (Hulme & Snowling 2009; Saksida et al., 2016; Vellutino et al., 2004).

Three additional studies have investigated the role of phonological deficits in deaf, dyslexic and typically developing children and identified different routes to reading in each group (Clark et al., 2016; Miller, 2005; 2007). Miller challenged the current view that a

phonological deficit underlies poor reading in dyslexia, and suggested an impairment in perceptual processing skills to be causal. With regard to the deaf students in all three studies, the authors argued that poor reading arose solely from language deficits or lack of access to early language. The deaf children in these studies were all sign language users. To the best of our knowledge, no previous study has taken a phonological deficit approach when comparing oral deaf children with hearing dyslexics.

In this paper, we compare the performance of oral deaf children and hearing children with a history of dyslexia on a range of literacy and phonological tasks. By exploring similarities and differences in each group, our study informs understanding of the reading profiles of different types of struggling readers, with implications for reading interventions. Our focus is on decoding skills, and the extent to which the SVR, used extensively to explain dyslexia in hearing children, also applies to oral deaf readers.

In the current study, we compared our two groups of participants on a battery of tasks that tap into the four key reading-related skills investigated by Caravolas et al., (2012) with hearing children: letter-sound knowledge, phonological or phoneme awareness, rapid automatized naming (RAN) and verbal short term memory. Caravolas and colleagues found that the first three of these skills were consistent predictors of individual differences in reading and spelling development across different alphabetic orthographies. Although the authors failed to find a significant predictive role of verbal short-term memory across orthographies, we included it in the current study due to previous findings of its association with reading outcomes and dyslexia (Wagner & Torgesen, 1987).

Studies of deaf children are usually based on small numbers because of the low incidence of the condition. Furthermore, studies typically include heterogeneous samples with mixed patterns of communication (i.e., children who use spoken or sign language or both), and wide age ranges. The present study compares the performance of a large sample of same-age deaf children, all of whom communicate principally using spoken language, with a group

of children with a history of dyslexia towards the end of their primary education. By this stage, reading is expected to be in place and if not, teachers need to know how to support the further development of literacy.

We sought to answer the following research questions:

(1) Do oral deaf children and hearing children with a history of dyslexia differ in literacy outcomes and performance on standardised reading-related measures known to be sensitive to dyslexia in hearing children?

(2) Are there differences in the concurrent correlates and predictors of literacy between deaf and dyslexic readers?

Method

Ethical approval for the study was obtained from City, University of London's School of Health Research Ethics Committee. Signed consent was received from all deaf and hearing children, their parents and from the head teacher of each participant's school or unit as applicable.

Participants

Deaf participants

Seventy-nine severely-profoundly deaf children, whose preferred form of communication was spoken language, aged between 10 - 11 years were recruited throughout the UK from special schools and services for deaf children and mainstream primary schools. Ten children were subsequently excluded as their non-verbal performance was more than one standard deviation below the mean, to be in line with the distribution of non-verbal scores of the hearing participants (see below). The final sample comprised 69 oral deaf (OD) children with a mean age of 11:00 years (SD = 4.37 months, range: 10;01-12;03 years) of whom 40 were girls. As the total sample was collected across two years, this represents 11% of the

population of severe-profoundly deaf children in this age range in the UK. Although all types of educational establishments were targeted, participants came mainly from units for the hearing impaired ($n = 53$) and mainstream schools ($n = 16$).

All children were reported by teachers and/or parents to be severe-profoundly deaf from birth (levels defined by the British Society of Audiology, 2011) and to use spoken English as their dominant language. Forty-four children used cochlear implants and 25 used digital hearing aids. Information about age at first aiding was missing for a third of the sample. Of the remainder ($n = 45$), 29 (63%) had been aided at or before the age of one year, 13 (30%) were aided between the ages of 2-3 years and the remaining 3 children were first aided between 5-6 years of age. As previously reported (Herman, Kyle & Roy, 2015), there were no significant differences on any of our key measures between children with cochlear implants or those with digital aids and so they were not separately analysed.

The inclusion criteria were that each deaf child had attended an English-speaking school from Year 1 and had no co-occurring difficulties (such as a severe developmental or visual impairment) that would make participation in the assessment tasks difficult. School staff confirmed that pupils met these criteria.

Data were available on additional languages spoken by the child for 52 participants. Over two thirds of these children (69%, $n=36$) were monolingual and spoke English only, a proportion of the group had knowledge of BSL ($n = 8$) or other spoken languages ($n = 7$), and one child used SSE. No child's main communication preference was BSL. Data on the presence of other deaf family members were returned for 46 participants (67%). Nine (20%) participants had one or more deaf members in their immediate family and of these, one child had one deaf parent and one child had two deaf parents.

Children came from a range of backgrounds in terms of parental education and ethnicity. Data were available for 58 participants (73%), of whom 42 (72%) were White British or White European, 8 (14%) were Asian British or Asian Other, 6 (10%) were Black British or

Black Other, and 2 (3%) were indicated as Other. Data on maternal education were available for 45 participants (65%): 9 (20%) had no qualifications; 6 (13%) had vocational qualifications; 15 (33%) had qualifications to GCSE (UK state examinations taken at the age of 16 years); 4 (9%) to A level (UK state examinations taken at the age of 18), and 11 (24%) had university degrees.

Hearing participants

Twenty hearing children aged between 8 – 11 years with a history of dyslexia (HOD), who had received a diagnosis of dyslexia during their primary school years, were recruited in England through mainstream schools, specialist schools for dyslexia or via researchers who had recently worked with families of children with dyslexia. The mean age was 10:00 years (SD = 7.83 months, range: 8;09-11;03 years) and 10 were girls. The sample of children with a history of dyslexia was younger than the deaf sample and with a wider age range; however, these children were recruited as a reference group, matched for reading ability, rather than a control group. All but one were reported to have hearing levels within the normal range. Four children had a history of persistent glue ear that had been treated with grommets; one of these had mildly impaired hearing.

Fifteen children were monolingual English speakers, one child had another spoken language at home and data were missing for four participants. Data on ethnicity and maternal education were available for 18 participants (90%). Sixteen (88.9%) were White British or White European, one (5.6%) was Black British and one (5.6%) Other. The association between ethnicity and hearing status was not significant (Fisher's exact=5.77, $p=.82$), in contrast to the association between maternal educational qualifications and hearing status (Fisher's exact=9.97, $p=.03$). One (5.6%) mother had no qualifications, five (28%) had vocational qualifications, one (5.6%) had qualifications to GCSE and two (11%) to A level, and nine (50%) had degrees. A disproportionate number of mothers were educated to degree

level in comparison with mothers in the deaf group and fewer had no or minimal educational qualifications.

Matching deaf and hearing groups on reading age

The OD group were significantly older than the HOD group ($t(22.53)=6.31$, $p<.001$), but their reading level was functionally the same. Analysis revealed that the reading age equivalent scores according to their performance on the British Ability Scales II (BAS II, Elliott, Smith & McCulloch, 1996) single word reading test did not differ ($t(87)=-.37$, $p=.71$). Likewise the non-verbal skills of the OD and HOD groups did not differ significantly [$\text{Mean}_{\text{OD}}=47.94$, $\text{SD}_{\text{OD}}=18.39$, $\text{Mean}_{\text{HOD}}=51.8$, $\text{SD}_{\text{HOD}}=25.05$; $t(25.22)=-.64$, $p=.53$].

Materials

All participants completed an extensive test battery of standardised assessments of non-verbal intelligence, reading and spelling, speech and language skills, and reading-related skills including measures of phonological awareness, letter-sound knowledge, RAN and verbal short-term memory, many of which had not hitherto been used with deaf children.

Non-verbal measures: The Matrices and Pattern Construction subtests from the BAS II, (Elliott et al., 1996) were used to assess the distribution of non-verbal abilities in the samples. The non-verbal score used was the mean percentile score across the two subtests.

Reading and spelling: Two assessments of reading ability were administered. The Single Word Reading subtest from the BAS II (Elliott et al., 1996) was the main measure used to identify poor readers (i.e., children with scores outside the normal range, $<-1\text{SD}$). Nonword reading ability was measured using the Dyslexia Portfolio Nonword Reading test (Turner, 2008). This test contains 45 nonwords ranging from one to six syllables in length that children read aloud. Spelling ability was measured using the Spelling subtest from the BAS II.

Speech and language: Three measures of speech and language were used. The Speech Intelligibility Rating Scales (SIRS, Allen, Nikolopoulos, Dyar & O'Donoghue, 2001) were used to rate the intelligibility of each deaf child's speech based on a short conversation with the researcher using a five-point scale. The Test of Child Speechreading (ToCS, Kyle, Campbell, Mohammed, Coleman & MacSweeney, 2013) was used to assess speech reading skills, since they are known to be predictive of literacy skills in deaf children (Kyle & Harris, 2006; 2011; Kyle et al., 2016). ToCS is a video-to-picture matching test that measures speech reading of single words, sentences and short stories and has norms for deaf and hearing children.

The Expressive One Word Picture Vocabulary Test (EOWPVT, Brownell, 2000) was used to measure oral vocabulary, which is known to impact on word recognition. This is a picture based naming task standardised on a US hearing population that has been used successfully with deaf children in the US (Yoshinaga-Itano, 2006) and in the UK (Johnson & Goswami, 2010; Kyle et al., 2016). In accordance with Johnson and Goswami (2010), four changes were made to test items to make them more appropriate for use in the UK: the item 'raccoon' was changed to 'badger'; the map of the US was replaced by a map of the UK; and the images for 'prescription' and 'windmill' were replaced by pictures that would be more easily recognised by British children.

Reading-related skills: Four subtests from the Phonological Assessment Battery (PhAB, Frederickson, Frith & Reason, 1997) were administered: Spoonerisms, Naming Speed, Fluency and Rhyme Awareness. In addition, three subtests from the Dyslexia Portfolio (Turner, 2008) were administered: Phoneme Deletion, Recall of Digits Forwards and Recall of Digits Backwards. These seven assessments measured phonological awareness at four different levels, RAN and verbal-short term memory.

The spoonerisms and phoneme deletion subtests measure manipulation of phonemes. The spoonerisms test requires children to transpose phonemes (e.g. 'cat' with a /f/ gives 'fat').

The phoneme deletion test instructs children to repeat words with parts omitted (e.g. say 'hedgehog' but don't say 'hog'; say 'fishy' but don't say /f/). Although demanding tests, good oral deaf readers have previously been shown to manage them well (Gravenstede & Roy, 2009). The rhyme awareness subtest requires children to identify two words that rhyme from a choice of three. The fluency subtest assesses phonological retrieval from long-term memory using either rhyme or alliteration and requires the child to name as many items as possible in 30 seconds from each category. This task has been shown to be a valid measure for deaf children (ibid.) and is discriminating in the diagnosis of dyslexia in the hearing population (Frith, Llanderl & Frith, 1995), with some evidence from a single case study of a signing deaf child with dyslexia (Herman & Roy, 2015).

The Naming Speed test measures RAN for pictures and digits using line drawings of everyday objects and numbers as stimuli. Although RAN is highly related to reading ability in the hearing population and sensitive to dyslexia, evidence from the deaf population of its relation to reading is mixed (Dyer et al., 2003; Gravenstede & Roy, 2009; Spencer & Tomblin, 2009). The raw score is time (seconds) to name the random sequence of 50 stimuli, accordingly and in contrast to the standard score, higher raw scores are indicative of poorer RAN performance. Verbal short term memory was assessed through recall of digits forwards and digits backwards subtests, where children are required to repeat number sequences of increasing length either forwards or backwards, and recalling familiar sequences, i.e., the days of the week and months of the year. The Letter-Sound Knowledge subtest from the YARC (Snowling et al., 2009) was used to measure children's knowledge of letters and sounds. This test requires children to say the sound associated with written letters. The extended version was used that includes all 26 letters and 6 digraphs which range from easy to more difficult (maximum score=32).

All tests requiring verbal responses (Single Word Reading, Nonword Reading, Speech Intelligibility Rating Scales, EOWPVT, Spoonerisms, Naming Speed, Fluency, Phoneme

Deletion, Recall of Digits Forwards, Recall of Digits Backwards, Letter-Sound Knowledge and familiar sequences) were checked for reliability.

Teacher and parent questionnaires were used to gather background information such as parental education, occupation, ethnicity and geographical location. Parents of OD children were additionally asked to provide information on their child's hearing including type of amplification and age when first used, and the duration of cochlear implant use where applicable.

Procedure

Children were tested over three or four sessions within one week in a quiet space in their school. Some of the HOD participants were assessed in either a quiet room in their home or at a designated testing centre. The order of testing was pre-determined and designed to vary task demands to maintain children's interest throughout each session (see Table 1). The first session started with non-verbal measures that included manipulation of equipment, then moved to a laptop based test of speechreading. The next session started with a paper based vocabulary test, followed by a series of short literacy tests. The remainder of the literacy tests, the phonological measures and sequencing were completed in a third and in some cases a fourth testing session.

Insert Table 1 here

All participants were assessed by a research assistant who was a qualified speech and language therapist, experienced in working with deaf children. In all cases, the researcher sat opposite children to ensure full access to speech reading cues in a quiet, well lit room, with hearing aids and cochlear implants fully functional. Tasks were administered as stipulated by the test manuals for both OD and HOD participants. No amendments were made to the

administration of the tests for the OD children, although the assessor always made it clear that the test instructions could, if necessary, be presented in BSL or SSE to ensure children knew what was required of them in order to complete the task. None of the OD participants requested communication support to understand the nature of the task, although additional explanation using writing was helpful to explain the phoneme manipulation tasks. For some OD children, extra practice items were included. At no point did the assessor fingerspell any of the words or present any of the contextual sentences in BSL or SSE.

Reliability

As many of the tests had not previously been used with deaf children and because of the impact of deafness on speech intelligibility, all tests requiring a verbal response (i.e., all except for non-verbal, spelling and speechreading measures) were audio and video recorded to check online scoring for accuracy and to investigate inter-rater reliability. A second rater, a qualified speech and language therapist, independently scored 10% of the data from the OD group. Reliability was high for all measures (above 0.7).

A further 10% of children were retested within a month to investigate test-retest reliability across all measures involving verbal responses. Retest reliability as measured by single measure intraclass correlations was overall found to be high, ranging from 0.89 to 1.00, the exception being the rhyme fluency test (0.67).

Results

Raw scores were used in all analyses, although standard scores are also presented to provide an indication of the groups' relative standing compared with their aged matched hearing peers (see Table 2). Using raw scores is beneficial as they are more discriminating at the low end of distributions for children performing at floor and achieving the minimum standard scores possible on any one test. In addition, it also partly addresses problems of

reliability associated with outdated norms on some tests used, for example, the PhAB (Frederickson et al., 1997).

Group differences in literacy, vocabulary and reading-related skills

Table 2 shows the means and standard deviations of standard and raw scores for measures of reading and spelling, expressive vocabulary, reading-related skills including measures of short term verbal memory, RAN, phonological awareness, and letter-sound knowledge and additional sequencing and speech measures, together with the effect size (d) of the group differences and their significance levels for the OD and HOD groups. Following Cohen (1988), $d=0.2$ is considered a ‘small’ effect size, $d=0.5$, a ‘medium’ effect size and $d=0.8$ a ‘large’ effect size. All standard scores had a mean of 100 and SD of 15.

A series of independent t-tests using Bonferroni correction for multiple comparisons (reducing significance level to $p<.003$) were used to evaluate the significance of group differences in mean scores. Group differences that remained significant after correcting for multiple comparisons are presented in bold.

Insert Table 2 here

As can be seen in Table 2 and might be expected from the word reading age equivalent scores, there were minimal differences between the OD and HOD groups’ word and nonword reading raw scores, and these were non-significant. The OD group achieved higher spelling raw scores but the between group difference was reduced to non-significance when the correction for multiple comparisons had been applied.

Overall the raw scores of the two groups were strikingly similar and effect sizes were small, with some exceptions. As can be seen in Table 2, the between group differences on measures of verbal short term memory, RAN for digits, phonological awareness

(spoonerisms), rhyme awareness and sequencing were small and non-significant. The oral deaf group had significantly higher scores on RAN for pictures and speech reading, both with large effect sizes. In contrast, between group differences in expressive vocabulary, letter-sound knowledge and rhyme fluency raw scores all favoured the younger HOD group. Once again, effect sizes were large. The group difference in phoneme deletion with a medium effect size, was reduced to non-significance once the Bonferroni correction for multiple comparisons was applied.

The OD children's letter-sound knowledge was particularly poor: 80% of the HOD group had ceiling scores of 30 or more compared with 16% of the OD group, and 15% of the HOD group achieved the maximum score of 32, compared with only 3% of the OD group. A substantial proportion of the OD group (43.5%) had scores below 27, the lowest score gained by only one child in the HOD group. Scattergrams of letter-sound knowledge scores against single word reading and nonword reading standard scores revealed that the nine OD children with very low letter-sound knowledge scores (<22) had below average word/nonword reading scores, with the exception of one child who had a single word reading standard score of 88. Whilst children with literacy scores in the average range had letter-sound knowledge scores at or above this score, so also did equal numbers of children with below average literacy scores, suggesting letter-sound knowledge is associated with but not sufficient for reading in the oral deaf children in our study.

In general, the standard scores tell a similar story to the raw scores. The literacy scores for both groups were between 0.5 – 0.75 SDs below the expected population mean of 100 (with a SD of 15). Both groups had a reading age (based on single word reading) of 9 years, although the OD group were older at a mean chronological age of 11:00 years, compared to the HOD group, who had a mean chronological age of 10:00 years. Taking the normative range as 1 SD above or below the mean, only expressive vocabulary, forward digit span and phoneme deletion mean standard scores in the OD group fell below this range (see Table 2).

From Table 2 and noted above it looks as if both groups were particularly challenged by the phoneme deletion task in comparison to their performance on other phonological awareness measures. However, a more parsimonious and cautious explanation might be related to the tests used. The phoneme deletion task was taken from a battery with norms established 10 years later than the PhAB battery which the other tasks were drawn from, a period marked by major changes in educational practice in the teaching of reading in the UK that has prioritised the role of phonics. On the other hand, notwithstanding concerns about the recency of norms, the superior performance of the OD group on the RAN tasks compared with their performance on other reading-related measures is of interest.

So despite apparent similarities in reading and spelling skills and a number of the reading-related measures, the profiles of underlying skills for the two groups differed in certain respects. However, a crucial question is the extent to which these similarities and differences were associated with literacy outcomes. To address this question, we considered correlations between reading and spelling outcomes and reading-related measures and expressive vocabulary.

Before turning to these results, we consider briefly the impact of two child/demographic factors that may affect children's performance. These factors are children's use of additional languages including BSL and number of deaf family members. A series of ANOVAs were run taking additional languages as the between factor with three levels (English only, BSL, and at least one additional language), taking the subsample of 52 children with data on this measure. These revealed no significant effect of additional languages; children with BSL or other languages did not differ from those with English only on any of our literacy or other measures included in Table 2. This applied to analyses where the one child with SSE was assigned to the BSL group ($n=9$) or other additional language(s) group ($n=8$). Secondly, a series of independent t tests were run to compare children from hearing families ($n=37$) with children from families where at least one member of the family was deaf

($n=9$). Once again most of the results were non-significant, with three exceptions: naming speed for pictures and backwards digit span favouring children from deaf families ($p=.004$, $p=.007$) and sequencing months favouring children from hearing families ($p=.006$).

Associations between literacy outcomes and reading-related measures and expressive vocabulary.

Associations between the three literacy measures (word reading, nonword reading and spelling) and reading-related measures and vocabulary were undertaken using Pearson correlations, taking each group separately. Once again, positive associations between age and the outcome measures within the two groups were relatively infrequent, but age was partialled out as a precautionary measure.

Insert Table 3 here

Table 3 shows the correlations between raw scores on word and nonword reading and spelling, and raw scores on the reading-related measures for the OD and HOD groups taking age as a covariate. Size and significance levels of the correlations are presented. Group differences in the size of the correlations were tested using the Fisher r to z transformations.

As can be seen, overall the correlations between measures in both groups were broadly similar. Strong associations between the three literacy measures and expressive vocabulary and measures of phonological awareness (phoneme deletion and spoonerisms) were evident in the OD group, and to a somewhat lesser extent in the HOD group. Differences between the correlations reached significance for the relationships between phoneme deletion and nonword reading and phoneme deletion and spelling ($Z = 2.32$, $p=.02$ and $Z=1.94$, $p=.05$ respectively), and borderline significance for the correlations between expressive vocabulary and word reading and expressive vocabulary and spelling ($Z=1.76$, $p=0.08$; $Z=1.85$, $p=0.06$).

Likewise, associations between rhyme fluency and nonword reading were significantly higher in the OD group compared with the HOD group ($Z=1.98$, $p=0.05$), and the higher associations between rhyme fluency and word reading and speech reading and nonword reading in the OD group were of borderline significance ($Z=1.9$, $p=0.06$; $Z=1.77$, $p=0.08$). The between group differences in correlations between spelling and rhyme fluency, and spelling and speech reading fell short of significance. In contrast, associations between verbal short term memory, RAN and letter-sound knowledge and literacy outcomes for both groups were similar. For both groups, letter-sound knowledge correlated most highly with nonword reading, and correlations with RAN for pictures and spelling were somewhat stronger in the OD group but not significantly so.

However, if a Bonferroni correction taking account of *all* between group differences in correlations for each literacy outcome measure is applied ($p=.004$), none of the above group differences would emerge as significant.

The strong correlation between word reading and expressive vocabulary raw scores is shown in the scattergram of these scores for both groups (see Figure 1). The Y reference lines represent the mean expressive vocabulary raw scores for each group. As can be seen in Figure 1, the strong association between low word reading scores and weak expressive vocabulary in the OD group was less evident in the HOD group. All OD children with low reading scores had low expressive vocabulary scores, as did some of the HOD with low reading scores. On the other hand, children in both groups at the top end of the distribution of word reading scores had expressive vocabulary scores at or above their group means.

Insert Figure 1 here

Given the strength of the correlations between expressive vocabulary and literacy scores and the known association between vocabulary and phonological processing skills in

hearing (Walley, Metsala & Garlock, 2003) and deaf children (Lee, Yim & Sim, 2012), the correlations were re-run taking vocabulary as a covariate to investigate the impact of reading-related skills on outcome measures once the effect of vocabulary had been taken into account.

Table 4 shows the correlations between raw scores on word reading, nonword reading and spelling and raw scores on the reading-related measures for the OD and HOD groups, taking age and expressive vocabulary as covariates. Similar to Table 3, the size and significance levels of the correlations are presented. Group differences in the size of the correlations were tested using the Fisher r to z transformations.

Insert Table 4 here

Overall, the effect of taking expressive vocabulary as a covariate was more marked in the OD group, with less impact on the r values of the HOD group. The correlations between verbal short term memory and word and nonword reading in the OD group were reduced to non-significance. There was minimal impact on the associations between literacy outcomes and RAN for pictures or digits. In the case of spelling, the enhanced correlation with RAN for digits in the OD group meant the between group difference in correlations reached borderline significance ($Z=-1.83$, $p=0.07$). Likewise, the impact on associations between letter-sound knowledge and literacy outcomes was minimal. In contrast, taking vocabulary into account reduced the size of the correlations between the phonological awareness measures and literacy outcomes, with the exception of alliteration fluency. The group difference in r values for spelling and phoneme deletion was reduced to non-significance but the difference for nonword reading and phoneme deletion remained significant ($Z=2.02$, $p=0.04$), but would be reduced to non-significance if the correction for multiple comparisons was applied. In contrast, the significantly higher correlations found previously between rhyme fluency and word/nonword reading in the OD group were accounted for by vocabulary, and the

associations between speech reading and literacy outcomes were reduced. Correlations between literacy outcome measures and non-verbal ability were reduced to non-significance and the size of the group difference in correlations for nonword reading increased and became of borderline significance ($Z=-1.7$, $p=.09$).

In summary, controlling for vocabulary had minimal impact on the associations found between literacy outcomes and RAN or letter-sound knowledge. Forward digit span, a measure of phonological short term memory, remained a significant predictor of reading in the HOD group only. Measures of phonological awareness, particularly those involving phonemic awareness and manipulation (phoneme deletion and spoonerisms) remained significant predictors for the OD group, albeit with smaller correlations. With regards to sequencing in the OD group, the association with nonword reading was no longer significant. Overall, spoonerisms emerged as a somewhat stronger predictor than phoneme deletion in the HOD group, and the correlation between spoonerisms and single word reading was the only one to remain significant with vocabulary controlled. Whereas small sample size in the HOD group played a part in reducing the significance levels of correlations between phonological awareness measures and literacy outcomes with vocabulary controlled, this could not explain the surprising lack of an association found between phoneme deletion and nonword reading in the HOD group that was even more striking with vocabulary controlled. Finally, we look at the relative strengths of these significant predictors for word reading, nonword reading and spelling for the oral deaf children.

Key concurrent predictors of decoding skills in the OD group

The extent to which vocabulary and reading-related skills predicted word reading, nonword reading and spelling in the OD group was explored further in a series of linear multiple regression analyses. As the sample size precluded entering all significant correlates simultaneously into the models, the following approach was adopted. In all cases, age was

entered first as a covariate and the three strongest predictors according to their r values (see Table 5) were then entered simultaneously. All remaining significant correlates were then entered, taking one at a time. All linear regression analyses were supported by robust statistics and bootstrapping. Preliminary checks of the assumptions underlying linear regressions revealed a significant outlier in the word reading analysis. This was due to an extremely low sequencing score (only four months were presented in the correct order). This child was removed from the word reading analysis.

Insert Table 5 here

Table 5 shows the standardised (β) and unstandardized (B) coefficients, the standard error, significance values and the 95% confidence intervals for B with and without bootstrapping and squared semipartial correlations (sr_i^2) for predictors of (a) word reading, (b) nonword reading, and (c) spelling. sr_i^2 provides a measure of the unique variance accounted for by each predictor in the models and the total amount of variance explained by unique variance and shared variance. As can be seen in Table 5, all three models were highly significant, accounting for between 71-79% of the variance in the outcome variables. It is striking in all three models that shared variance accounts for the majority of total variance explained, more so for word reading and less so for spelling, but in all cases the amount was substantial.

Not surprisingly, expressive vocabulary was a strong and unique predictor in all models, explaining the highest amount of unique variance compared with the unique contribution of other predictors in all models, albeit a relatively smaller amount for nonword reading. Phoneme deletion also emerged as a unique predictor of all three outcome measures, explaining as did spoonerisms with the exception of spelling, small but significant amounts of variance in all models. RAN for digits was a strong and unique predictor of spelling,

accounting for the same amount of unique variance explained as vocabulary and also added significantly to the prediction of nonword reading. Letter-sound knowledge emerged as a significant predictor of nonword reading only, accounting for a small amount of additional variance. Overall the results were supported by the results of the bootstrapping analyses with the exception of sequencing for word reading where the small contribution to the final model was reduced to non-significance. None of the remaining significant reading-related correlates added significantly to the models.

We repeated the regression analyses taking expressive vocabulary as the outcome variable and the literacy variables [single word reading (model A), non-word reading (model B) and spelling (model C)] as predictors, entering the same predictors as before, with age entered first as a covariate. Analyses were again supported by robust statistics and bootstrapping.

R square and adjusted R square for model A were .7 and .67 respectively, compared with .8 and .79 respectively for model A (see Table 5) when word reading was the outcome variable and expressive vocabulary the predictor. For model A, the amount of unique variability that was explained was 18% and the amount of shared was 52%. Word reading was the only predictor that added significantly to the model $p < .001$, $p(\text{BS}) = .001$; none of the other predictors added significantly. For model B, taking expressive vocabulary as the outcome measure and including non-word reading as a predictor, R square was .62 and adjusted R square was .58 compared with .73, and .71 for model B (see Table 5). In model B, the amount of unique variance explained was 18.6% and the amount of shared was 43.4%. Nonword reading as a predictor added significantly $p = .001$, $p(\text{BS}) = .003$, accounting for 7.5% unique variance, but so did most of the other predictors: RAN digits $p = .12$, $p(\text{BS}) = .09$, 1.6% unique variance; letter-sound knowledge $p = .01$, $p(\text{BS}) = .004$, 4% unique variance; spoonerisms $p = .02$, $p(\text{BS}) = .002$, and phoneme deletion (borderline significance) $p = .07$, $p(\text{BS}) = .07$, 1.7% unique variance. For model C, taking expressive vocabulary as the outcome measure and

including spelling as a predictor, R square was .68 and adjusted R square was .66 compared with .81, and .79 when spelling was the outcome measure (see Table 5). In model C, the amount of unique variance shared was 36.2% and shared variance was 31.8% spelling as a predictor added significantly $p < .001$, $p(\text{BS}) = .001$, accounting for 24.7% of unique variance, as did RAN digits $p < .001$, $p(\text{BS}) = .001$, 10% unique variance with phoneme deletion $p = .09$, $p(\text{BS}) = .1$, 1.5% unique variance.

Together these regressions analyses show that strong associations between literacy measures and expressive vocabulary held for both directions of the relationships, with somewhat more variance explained when expressive vocabulary was acting as predictor for literacy than vice versa when literacy predicted expressive vocabulary. As in the previous analyses, taking word and nonword reading as predictors shared variability accounted for a greater proportion of the total variance compared with the unique variability of predictors taken separately. The scattergram (Figure 1 above) illustrates the mutual strength of this association for the OD group: unlike the HOD group, none of the OD struggling readers had reasonable vocabulary that typifies the classic dyslexic profile.

Discussion

This study set out to compare the profiles of two groups of children with reading difficulties with different developmental pathways who have not hitherto been compared: oral deaf children and hearing children with a history of dyslexia. In comparison with other studies we were successful in recruiting a uniquely large sample of same age deaf children, a group that is difficult to access, all using the same form of communication: spoken English. The fact that half of the OD group were reading at age level is encouraging, indicating that poor reading is not an inevitable outcome for deaf children, and points to the variability that has been reported elsewhere within this group (Mayer & Trezek, 2017). For the current group of oral deaf children, the performance of a small sub-group who used BSL as an additional

language did not differ from those who used spoken English only on any measure. In contrast, the dyslexic sample was surprisingly difficult to recruit. It was not possible to match children for age, but the groups were matched for reading ability, and our study focused in the main on the relationships between scores in each group. We now return to our research questions.

Do oral deaf children and hearing children with a history of dyslexia differ in literacy outcomes and performance on standardised reading-related measures known to be sensitive to dyslexia in hearing children?

Addressing our first research question, despite having distinct aetiologies, several similarities in outcomes were observed between the two groups. In addition to being matched on the word reading measure, our analyses revealed that deaf and dyslexic children's standard scores on nonword reading and spelling were also strikingly similar, falling within the average range, albeit at the lower end. Nonword reading was the weakest in both groups, being nearly 1 SD below the mean in both cases. Although some researchers have questioned the value of nonword reading in identifying struggling readers (Thompson & Johnston, 2000), nonword reading is frequently used as a measure of decoding and these findings are in keeping with previous research on dyslexic children (Elbro & Jensen, 2005; Vellutino et al., 2004).

Although the HOD group mean literacy scores fell in the normal range, they were arguably higher than would be expected from a group of children with dyslexia; nonetheless, similar to the OD group, there was variability in scores within the group. At least two factors may play a part in these higher than expected scores. First, all children had received interventions targeting their dyslexia following their diagnosis, therefore a number of children might be considered as 'compensated' dyslexics, with higher scores a direct consequence of the success of the interventions received (see below). However, some were still reading at a low level, pointing to the heterogeneity of the group and the disorder. Second, as a group their

mean literacy scores are strikingly similar to the mean scores reported for the ‘at-risk impaired’ group of children in Snowling and colleagues’ longitudinal study of familial dyslexia (Snowling, Gallagher & Frith, 2003; Snowling et al., 2007). The ‘at-risk impaired’ group had mean scores between .75 and -2SDs below the ‘at-risk unimpaired’ and the control groups aged 8 years and at follow-up aged 12-13 years. As the authors noted, their cut-off score taken to be 1SD below the control group mean WORD composite score of 114.49 (SD=14.0) could be seen as ‘lenient because children with this level of attainment would not normally be classified as having a reading disability’ (2003, p. 363). Like our HOD sample, their children were from families of relatively high socio-economic backgrounds. As the robust relation of socioeconomic status to reading outcomes is well established (Aikens & Barbarin, 2008), future research with larger samples should take SES differences into account.

Children with dyslexia typically struggle with reading-related tasks that tap phonological skills. However, with the possible exception of phoneme deletion, the phonological awareness scores of the HOD group were within 0.5 SD below the mean and higher than their reading, spelling and short term memory scores. This profile may be a direct consequence of the dyslexia support they have received since diagnosis, which typically targets phonological skills (see Duff & Clarke, 2011 for a review of interventions for dyslexia), together with the known specificity of effects on targeted skills (Hatcher, Hulme & Ellis, 1994). These elevated phonological scores, possibly the result of effective interventions, may have played a part in the striking lack of association found between phoneme deletion and nonword reading scores in the HOD group only. Inspection of a scattergram of phoneme deletion standard scores against nonword standard scores lent some support to this argument, with very high scorers on the phoneme deletion task not matched with exceptional nonword reading scores.

As others have found (see Clark et al., 2016), the most striking difference between the OD and HOD groups was the exceptionally weak vocabulary skills of the OD children. Our

findings for the OD group mirror and add to those of other researchers that have repeatedly shown deaf children to have vocabulary delays as a result of challenges in accessing spoken language from an early age (Lund, 2016). Whilst the majority of HOD children had vocabulary scores as expected, either in or above the average range, it was apparent that a minority of HOD poor readers had weak language skills, together with phonological deficits, a profile that typified the OD poor readers. These children may be considered at risk for poor reading comprehension in future (Bishop & Snowling, 2004; Kyle & Cain, 2015). In the group of poor OD readers, no children mirrored the classic dyslexic profile of weak phonological skills alongside good language skills. All deaf children who were poor readers displayed weak language, the so-called ‘garden variety’ type (Gough & Tunmer, 1986). Our study highlights the ‘heterogeneity among individuals who develop a specific disorder’ (Cicchetti & Rogosch, 1996, p. 597), observed in these groups of children sharing the same diagnosis but whose profiles of strengths and weaknesses in practice extend along a continuum (Rose, 2009).

While the OD participants scored somewhat lower than the HOD children on the phonological measures, the main group difference observed in raw scores (which are less affected by the normative changes), was in rhyme fluency. Subsequent analyses showed that performance on the rhyme fluency task was strongly associated with the exceptionally weak vocabulary knowledge of the OD group. Letter-sound knowledge was also significantly poorer in the OD readers, and emerged as a significant predictor of their nonword reading. Letter-sound knowledge is typically mastered at an early stage in the development of literacy, with the majority of the younger HOD group at ceiling on this task. The fact that so few OD children had achieved full letter-sound knowledge competency at the end of their primary education, given its key role in decoding skills for hearing (Shapiro, Carroll, & Solity, 2013) and deaf children (Goldberg & Lederberg, 2015; Kyle & Harris, 2011), can be seen as cause for concern. However, for some able and precocious readers there is evidence that *full* letter-

sound knowledge is not essential to achieve reading competence (Thompson, 2014; Thompson, Connelly, Fletcher-Flinn, & Hodson, 2009). On the other hand, it remains an empirical question whether children who find reading a challenging and difficult skill to acquire need component skills such as letter-sound knowledge to be taught explicitly.

Recall of digits forwards, used as a measure of phonological short-term memory, also emerged as particularly challenging for the OD group. Although they also achieved low scores on the recall of digits backwards task, their mean score on this remained within the normal range and was substantially higher than their forward digit span scores. Other researchers have reported poor digit span performance in deaf children, for both forwards and backwards tasks (Burkholder & Pisoni, 2003; Pisoni & Cleary, 2003; Pisoni, Kronenberger, Roman & Geers, 2011), attributing it to slower verbal processing, and showing correlations with speech and language outcomes in adolescence for deaf young people with cochlear implants, including reading (Harris et al., 2013). Pisoni and colleagues suggested that for deaf children, digits forwards may be more difficult because it emphasizes the phonological loop of memory and sequential processing, both of which are at risk in deaf children, whereas digits backwards emphasizes other processes, such as central executive and attention, as well as immediate verbal memory and sequential processing (Pisoni et al., 2011).

Of all our reading-related measures, RAN was the one where the OD group as a whole displayed the expected distribution of scores and did not differ from the test norms, indicating intact skills. The OD group was better on the pictures task than digits. Furthermore, in a single case study of a signing deaf child with suspected dyslexia (Herman & Roy, 2015), lower scores were obtained for RAN for digits than pictures, similar to the pattern found among hearing children with dyslexia (Savage & Frederickson, 2006; Semrud-Clikeman, Guy, Griffin, & Hynd, 2000).

A final area of interest was the degree to which the groups differed in their speechreading performance. The speechreading task was normed on deaf and hearing children

(Kyle et al., 2013) and it was expected that the OD group would achieve scores within the normal range. For our HOD participants, as in previous studies with dyslexic children (de Gelder & Vroomen, 1998) and adults (Mohammed, Campbell, MacSweeney, Barry & Coleman, 2006), the speechreading task was challenging.

Are there differences in the concurrent correlates and predictors of literacy between deaf and dyslexic readers?

Turning to our second research question, there were some striking similarities and differences in the concurrent correlates and predictors of literacy between the deaf and hearing groups. Importantly, we have shown that, like hearing children, phonological measures involving direct manipulation of phonemes (phoneme deletion, spoonerisms) and letter-sound knowledge play a role in OD children's reading, with RAN for digits important for nonword reading and spelling. Almost all OD children had speech that was intelligible, with only three presenting with speech that was difficult to understand in spontaneous conversation. Yet despite this, only just over half (52%) were reading age appropriately, therefore speech intelligibility alone is not predictive of good reading in OD children (see also Harris & Moreno, 2006; Leybaert & Alegria, 1995). In the current study, the degree of speech intelligibility accounted for between a quarter to a third of the variance in the literacy outcome measures, although it was not significant once vocabulary and phonological measures were taken into account. We have shown previously that speech intelligibility is related to the use of the phonological route in word spelling and reading, suggesting that it may be more indicative of underlying strategies rather than a key factor in literacy outcomes (Roy, Shergold, Kyle & Herman, 2015).

Furthermore, the correlations between vocabulary and literacy outcomes were all strong in the OD group compared with 'moderate' associations found in the HOD group. Our findings and those of others show that these vocabulary delays are strongly associated with

poor reading (Kyle & Harris, 2010; Kyle et al., 2016; Lund et al., 2016; Mayberry et al., 2011; Musselman, 2000). The key role of vocabulary in the OD group was also confirmed by our regression analyses, which identified the major contribution of vocabulary to all literacy outcomes in the OD group. However, equally striking, particularly in relation to single word and nonword reading was the overwhelming amount of total variance explained by *shared* as opposed to *unique* variance. In other words, whilst highlighting the importance of language for literacy, our evidence underpins the importance of recognising the *interdependence* of vocabulary with other phonological reading related skills for achieving literacy. The current study is correlational in nature which limits our ability to infer the direction of causality between vocabulary and literacy skills in oral deaf children. In order to determine the direction of the relationship, we ran the same regression models but with vocabulary as the outcome variable and the literacy variables as the predictors (sample size precluded the use of regression with the HOD children). For the OD children, vocabulary was a somewhat stronger predictor of all three literacy outcomes rather than vice versa, but there was clearly a lot of shared variance and it makes sense that the relationship is bidirectional (see Kyle, 2015).

The results also provide further support for the argument that a lack of early and complete language access is a large contributory factor to low levels of reading attainment in deaf populations (e.g. Clark et al., 2016). The results of two recent studies are particularly interesting in relation to this (Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012). Both of these studies found that deaf children with cochlear implants who had deaf parents and had early sign exposure had better language outcomes than deaf children with cochlear implants and hearing parents. The strong relationship typically observed between language and reading outcomes in deaf students would suggest that these children who had full and early access to *both* spoken and sign languages would be likely to go on to have better reading outcomes.

Vocabulary was a moderate predictor of reading in the HOD group, accounting for over a third of the variance in their word and nonword reading scores in line with previous research (Nation & Snowling, 2004; Ouellette & Beers, 2010; Song et al., 2015). Evidence from hearing children has shown that it is familiarity with the phonological forms of words rather than semantic knowledge that is key in the robust relationship found between word knowledge and reading aloud success (Nation & Cocksey, 2009). This evidence was drawn in the main from samples of typically developing children. The nature of the association between word knowledge and reading in children with weak vocabulary, including deaf children, struggling readers with co-occurring language problems, and children from low socioeconomic backgrounds with impoverished input (Roy & Chiat, 2013) warrants further investigation.

It was expected that speechreading would predict reading in the OD group, given other research findings of its role in deaf reading (Kyle et al., 2016; Kyle & Harris, 2010; Rodríguez-Ortiz, Saldaña, & Moreno-Perez, 2015). It was a consistent correlate of decoding literacy for our OD participants, but not once vocabulary and/or phonological skills were controlled for. This is in contrast to the studies of Kyle and colleagues (Harris et al., 2017; Kyle & Harris, 2010, Kyle et al., 2016), where speechreading was a significant predictor of reading ability in deaf children; however, the children in those studies were younger and from mixed language backgrounds, and phonological skills were not controlled for. The findings from the current study point to interventions for oral deaf children that focus on the development of vocabulary and phonological skills, with speechreading providing visual support.

There was no significant association between speechreading and reading ability in our HOD participants. This is in contrast to a previous study with dyslexic adults (Mohammed et al., 2006) in which they found that phonological skills accounted for the relationship between speechreading and reading ability. The lack of association in the current study may be due to

participants' improved phonological skills resulting from dyslexia support, which teaches phonological strategies and lessens the need to supplement phonological information with visual detail.

Our findings support those of Dyer et al. (2003) who explored the relationships between phonological tasks, RAN and reading in a group of 49 deaf teenagers. Similar to the current study, Dyer et al. failed to find a significant relationship between reading ability and picture-based RAN. However, the current study also included measures of nonword reading and spelling together with a digit-based RAN task, which tends to be a more consistent predictor of reading ability than picture-based RAN. We found that digit-based RAN was predictive of both spelling and nonword reading ability in the OD group, even after controlling for vocabulary and phonological skills, in line with evidence from English speaking hearing children (Stainthorp, Powell & Stuart, 2013). Like Bishop et al. (2009), we found the RAN measures were unrelated to vocabulary, a measure of expressive language. This finding, together with the fact that the poor phonological awareness skills of the OD group did not affect their capacity to access the familiar phonological forms involved in the RAN tasks, underscores the predictive value of RAN for digits in identifying dyslexia in OD children, where such a high proportion showed significant language problems (see Bishop et al., 2009 for comparable results in hearing children with language impairments).

A final area of difference between the groups was in the predictive role of digit span. Despite the OD children showing particular difficulties on the recall of digits forward task, once vocabulary was taken into account, our study found no association between digit span and literacy (with the exception of spelling) in the OD group, whereas it remained significant for the HOD group. This suggests the task may be tapping different skills in the two groups. One possibility is that in the absence of cues in the forward digit task, the OD children did not spontaneously rehearse, whereas they may visually recode verbal material in backwards span where their recall was substantially better. Backwards span as a measure of working memory

was not significantly associated with reading outcomes in either group once vocabulary was taken into account. Overall, in line with Caravolas et al.'s (2012) findings with hearing children, short term verbal memory as measured by forwards digit span did not emerge as useful predictor of literacy outcomes in our OD sample.

Implications of findings

The above results, along with the different pattern of speechreading associations in the two groups, suggest that although OD and HOD exhibit equifinality in their reading profiles in terms of decoding outcomes and associations with phonological skills, they have achieved phonological skills via different developmental pathways. OD children are likely to have developed their phonological skills in part through speechreading, or through the experience of reading itself, which has led to debate in the equivalence of different routes (see Kyle & Harris, 2010; Mayberry et al., 2011; McQuarrie & Parilla, 2009). The current results suggest that, although OD children's phonological representations integrate phonological information from different sources, they do not appear to spontaneously rehearse auditory or phonological material unless explicitly cued to do so, as is the case in backwards digit span. These results provide further insight into the complex nature of phonological skills and phonological decoding in oral deaf children.

Some recent research (Allen, 2015) has shown that fingerspelling can significantly support the development of letter skills in deaf children in native signing families when introduced in the home or in school settings (Haptonstall-Nykaza & Schick, 2007; Stone et al., 2015). Other studies have advocated for the use of Visual Phonics to support the acquisition of letter-sound knowledge (Trezek & Malmgren, 2005; Trezek & Wang, 2006). These approaches have generally been trialled with deaf children who communicate using sign. Their applicability to oral deaf children requires further investigation.

Our study represents a first step in exploring multifinality through inspection of the heterogeneous profiles found in each group (Cicchetti & Rogosch, 1996), although sample size constrained the kinds of analyses of sub-groups that could be carried out. While the majority of HOD children had good language skills, this was not true for all. Whereas that many were performing near to age level on many measures may be testament to the effectiveness of dyslexia interventions, not all achieved high scores, suggesting that more research is needed to identify which interventions work best with different profiles. In particular, dyslexic children with language difficulties are unlikely to make sufficient progress with interventions that focus solely on their phonological difficulties, as they additionally need support with language. Previous research with hearing children has indicated that interventions show effects for areas that are directly targeted, and that integrated interventions convey advantages for reading over interventions with a single focus (Hatcher et al., 1994; Lonigan, Purpura, Wilson, Walker & Clancy-Menchetti, 2013). For both groups of struggling readers, interventions are needed that address phonics and language skills and *all* areas of weakness within a fully integrated approach, rather than in the form of separate interventions delivered simultaneously. Our findings on the substantial role of *shared* variance in the OD group, and the interdependence of vocabulary and phonological skills further underscores this need for fully integrated interventions.

In the OD group, we identified profiles of good and poor readers. Among the good readers, a subset appear to be at risk because of poor language skills. Although reading at age level now, support to develop language skills is needed to prevent the plateau in literacy skills that has been so widely observed in this group (e.g. Allen, 1986; Qi & Mitchell, 2011). Of the poor deaf readers, future research could usefully explore whether some display a more dyslexic profile, for which current reading interventions may be appropriate. However, all poor deaf readers additionally require language support to underpin their literacy development.

Finally, we have established that it is possible to use this carefully selected range of tests effectively with oral deaf children. Beyond effectiveness, we have shown that performance on some measures, such as forward digit span, may not be directly comparable to hearing children and such results should be interpreted with caution. This finding will be of value to literacy specialists whose job it is to assess and offer support for children with reading difficulties in schools, some of whom are deaf.

Limitations

There are several limitations to the current study. Since it focused solely on oral deaf children, it carries implications mainly for this subgroup of deaf children, who nonetheless constitute the majority of the deaf population. Future research that includes signing deaf children is warranted as different findings may emerge, with consequences for intervention approach. Indeed, data collected using a similar test battery with a sample of same-age signing deaf children are the subject of a separate paper. Some of the children in the current study had exposure to BSL, yet their BSL skills were not evaluated, even though measures of sign language acquisition are increasingly available (Enns et al., 2016). Although these children did not use BSL as a primary form of communication or request it being used during their assessment, the extent to which their exposure to BSL influenced their literacy skill development is unknown.

Another limitation is the use of only a single measure of expressive vocabulary as a proxy for language. It is suggested that future studies include more measures of language, such as receptive vocabulary and higher order language skills. A further shortcoming, due partly to funding constraints, was a lack of information in the HOD group on age of diagnosis of dyslexia, frequency, duration and type of intervention(s) received, and profile of scores at the point of diagnosis. Such information would have permitted a more nuanced understanding

of the children's profiles. Furthermore, there was a lack of information about literacy instruction received by children in both groups.

Finally, although our study has established that it is possible to use this selected range of tests, developed for and standardised on hearing children, effectively with oral deaf children, this does not necessarily make it valid for this population. Investigations of the validity of these and other assessments when used with oral deaf children are warranted.

Conclusion

This study compared two groups of struggling readers, OD children and HOD children, using a range of literacy and reading-related measures developed for hearing children. Our analysis has deepened our understanding of reading profiles in each group and paves the way towards the use of latent profile analysis as an alternative methodology in future research with larger samples of participants.

Our findings show that letter-sound knowledge, measures of RAN and phonological skills, all of which are important in the field of dyslexia, are also informative in understanding deaf children's reading difficulties. By confirming the value of these key measures, literacy specialists in schools can be more confident in their approach to assessing deaf children's reading and reading-related skills. For practitioners, the knowledge that such measures can effectively identify deaf children at risk of reading difficulties is a first step towards providing appropriate support to consolidate early reading skills in these areas, in the same way as is provided to dyslexic children.

The OD participants achieved much lower scores than the HOD children on rhyme fluency, phoneme deletion and vocabulary knowledge and two of these skills, phoneme deletion and vocabulary, were found to be highly predictive of literacy outcomes. This highlights the key role of language for oral deaf children's reading, in combination with

phonological skills, where the directionality of effects can be difficult if not impossible to determine.

Our HOD sample was matched on reading age but was younger than the OD participants. They were recruited from a narrower geographic spread and differed in other background variables such as maternal education. In addition, all had benefited from specialist interventions with many attending schools specifically for children with dyslexia, thus they can be considered compensated to some degree. The outlook for these children is relatively optimistic. With ongoing specialist support and not yet at the end of primary school, they may be expected to improve their literacy skills before secondary transition. The same cannot be said of the OD participants, all of whom were at the end of primary education, at an age when competence in reading would be expected, and unlike the HOD children, none were benefitting from specialist literacy support.

The large body of research on childhood dyslexia has contributed to a better understanding of poor reading in this group of children, which in turn has informed the design of effective reading interventions (Duff & Clarke, 2011). It is equally important to implement and evaluate reading interventions to support deaf children's reading. Our work underscores the important role of language in the identification and remediation of reading difficulties, highlighted from research with hearing children (Catts, Fey, Weismer & Bridges, 2014). Reading interventions for deaf children need to specifically target deaf children's vocabulary deficits in addition to their weak phonological skills within a fully integrated approach. Language skills are important for the early stages of learning to read and essential for reading comprehension (Herman, Roy & Kyle, 2017).

Ideally, language interventions need to be offered at a much earlier stage before children embark on formal education to prevent the consequences of language deprivation impacting on literacy and academic achievement. Furthermore, interventions that incorporate sign language may be beneficial (Clark et al., 2016). Without adequate and targeted support,

our findings, along with others, indicate the serious challenges that deaf children will continue to face at school.

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