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Language Development and Impairment in Children with Mild to Moderate Sensorineural  
Hearing Loss

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This research was funded by an ESRC First Grants Award (RES-061-25-0440) awarded to Lorna F. Halliday.

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## Abstract

**Purpose:** To examine language development and factors related to language impairments in children with mild to moderate sensorineural hearing loss (MMHL).

**Method:** Ninety 8-16 year-old children (46 children with MMHL; 44 aged-matched controls) were administered a battery of standardized language assessments, including measures of phonological processing, receptive and expressive vocabulary and grammar, word and nonword reading, and parental report of communication skills. Group differences were examined after controlling for nonverbal ability.

**Results:** Children with MMHL performed as well as controls on receptive vocabulary and word and nonword reading. They also performed within normal limits, albeit significantly worse than controls on expressive vocabulary, and receptive and expressive grammar, and worse than both controls and standardized norms on phonological processing and parental report of communication skills. However, there was considerable variation in performance, with 26% showing evidence of clinically significant oral or written language impairments. Poor performance was not linked to severity of hearing loss nor age of diagnosis. Rather, outcomes were related to nonverbal ability, maternal education, and presence/absence of family history of language problems.

**Conclusions:** Clinically significant language impairments are not an inevitable consequence of MMHL. Risk factors appear to include lower maternal education and family history of language problems, whereas nonverbal ability may constitute a protective factor.

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Sensorineural hearing loss (SNHL) is a permanent hearing impairment caused by a defect in the cochlea or auditory nerve (Moore, 2007). Individuals with mild or moderate losses have average hearing thresholds of between 21 and 70 dB HL (mild: 21-40 dB HL; moderate: 41-70 dB HL; British Society of Audiology, 2011). As such, they have residual hearing that is useful without the assistance of hearing devices such as hearing aids or cochlear implants, albeit hearing that is degraded. Recent estimates suggest that globally, around 6.2% of children aged 5 to 14 years have a mild hearing loss, compared with 0.2-1.7% who have moderate losses (although rates are much lower in developed compared to developing regions; Stevens, Flaxman, Brunskill, Mascarenhas, Mathers, & Finucane, 2013). However, despite the high prevalence of this condition, relatively little is known about the outcomes of children with mild or moderate SNHL (MMHL). The current study examines individual differences in the language development of children with MMHL.

The language environment experienced by children growing up with MMHL differs substantially from that of children with normal hearing. In addition to raising hearing thresholds, SNHL leads to a broadening of auditory filters and changes the way in which sounds are processed (see Moore, 2007, for a review). The consequence of this is that children with even mild or moderate levels of SNHL are likely to experience a speech signal that is distorted or degraded, as well as frequently being quieter, and with important acoustic cues near or below threshold. To some extent, the introduction of universal newborn hearing screening programmes in many developed countries has gone some way towards addressing this, by identifying SNHL in infancy in some children. However, in the UK this programme is not designed to detect mild levels of hearing loss (Bamford, Uus, & Davis, 2005), meaning that many children born with mild congenital SNHL will not be detected until later in childhood (Watkin & Baldwin, 2011). Moreover, even when SNHL is identified, the introduction of hearing devices such as hearing aids and FM systems only goes so far towards

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addressing the problem. Typically, whilst these devices boost the intensity of the signal and compress variations in level, they do not rectify many of the perceptual consequences of SNHL (Moore, 2007). Compliance can also be a problem in that many children do not always use their hearing aids (Fitzpatrick, Durieux-Smith, & Whittingham, 2010; Walker, Holte, McCreery, Spratford, Page, & Moeller, 2015). Consequently, children with even mild to moderate levels of SNHL are faced with having to learn an oral language from an auditory signal that is partial, distorted, and/or degraded.

How well children with MMHL fare learning language varies considerably. In general, however, they have much success. Children with MMHL will acquire their oral language spontaneously, and most do not rely on sign language or use visually coded systems such as cued speech, although they may rely more on speech-reading to bolster their language acquisition. These days, the vast majority of children with MMHL are educated in mainstream schools, although many still have additional support. However, despite this, studies have typically shown that children with MMHL perform more poorly than their normally hearing peers on standardised and experimental measures of language, including vocabulary and word learning, morphology and syntax, and phonology and reading, although these studies have also tended to identify marked individual variability in performance (for reviews, see Lederberg, Schick, & Spencer, 2013; Moeller, Tomblin, Yoshinago-Itano, Connor, & Jerger, 2007). A brief summary of this literature is provided here.

There are now several studies that have examined the vocabulary and/or novel word learning skills of children with SNHL, including those with MMHL. In general, these studies have reported evidence for delays and/or deviancies in the development of receptive and expressive vocabularies of children with MMHL relative to normally hearing controls (Blamey et al. 2001; Davis, Elfenbein, Schum, & Bentler, 1986; Davis, Shepard, Stelmachowicz, & Gorga, 1981; Fitzpatrick, Crawford, Ni, & Durieux-Smith, 2011;

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Gilbertson & Kamhi, 1995; Kiese-Himmel & Reeh, 2006; Mayne, Yoshinaga-Itano, & Sedey, 1998; Mayne, Yoshinaga-Itano, Sedey, & Carey, 1998; Pittman, Lewis, Hoover, & Stelmachowicz, 2005; Wake, Hughes, Poulakis, Collins, & Rickards, 2004; Wake, Poulakis, Hughes, Carey-Sargeant, & Rickards, 2005; c.f. Briscoe, Bishop, & Norbury, 2001; Delage & Tuller, 2007; Halliday & Bishop, 2005; Hansson, Forsberg, Lofqvist, Maki-Torkko, & Sahlen, 2004). Moreover, delays and difficulties have been demonstrated in the word learning skills of children with mild to severe SNHL (Gilbertson & Kamhi, 1995; Lederberg, Prezbindowski, & Spencer, 2000; Pittman et al., 2005; Stelmachowicz, Pittman, Hoover, & Lewis, 2004; c.f. Hansson et al., 2004), and these have in general been linked to the smaller vocabulary sizes of this group relative to their age-matched normally hearing peers (Lederberg et al., 2000; Pittman et al., 2005; Stelmachowicz et al., 2004). However, those that have looked have often reported evidence for substantial individual differences in the vocabulary and word learning skills of these children (Gilbertson & Kamhi, 1995; Kiese-Himmel & Reeh, 2006; Wolgemuth, Kamhi, & Lee, 1998). For instance, these studies have suggested that only around 12-50% of a given sample of children with MMHL are likely to have vocabularies and/or word learning abilities that are outside normal limits (Delage & Tuller, 2007; Gilbertson & Kamhi, 1995; Kiese-Himmel & Reeh, 2006; Sikora & Plapinger, 1994).

The factors underlying these individual differences are not well understood. However, it is clear that severity of hearing loss is not the whole story. Although some of these studies have found that vocabulary and/or word learning skills get worse with poorer hearing thresholds (Davis et al., 1986; Wake et al., 2004; Wake et al., 2005), others have not (Blamey et al., 2001; Gilbertson & Kamhi, 1995; Mayne et al., 1998a, 1998b; Sikora & Plapinger, 1994), and several have shown that even children with the mildest levels of hearing loss as a group show delays in vocabulary development (Davis et al., 1986, 1981; Wake et al., 2004).

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Others have demonstrated the importance of age of detection in determining outcomes (Kennedy et al., 2006; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998; c.f. Wake et al., 2005). Moreover, age and/or quality or quantity of intervention (Mayne et al., 1998b; Moeller, McCleary, Putman, Tyler-Krings, Hoover, & Stelmachowicz, 2010; Nittrouer & Burton, 2003; Tomblin, Harrison, Ambrose, Walker, Oleson, & Moeller, 2015; Walker et al., 2015; c.f. Mayne et al., 1998a; Pittman et al., 2005), along with family involvement (Moeller, 2000; Moeller et al., 2010), have been shown to predict outcomes. At least two studies have identified a role for nonverbal ability in predicting both receptive and expressive vocabulary development (Mayne et al., 1998a, 1998b). It has also been argued that those children with SNHL who perform poorly on these language tasks may have an additional, undiagnosed, Language Disorder (Gilbertson & Kamhi, 1995), although there is currently no evidence to support this claim. Finally, it is possible that whilst children with MMHL may get off to a slow start with their vocabulary development, many of them subsequently catch up with their peers. For instance, Kiese-Himmel and Reeh (2006) found that of the 16 2-4 year-old children with MMHL that they followed longitudinally, a quarter showed expressive vocabulary abilities commensurate with their normally hearing peers after 9 months. It is also noteworthy that of the four existing studies of children with MMHL that included teenagers in their sample, three either found no significant group differences in vocabulary skills (Halliday & Bishop, 2005; Hansson, Sahlen, & Maki-Torkko, 2007) or reported that only a small percentage (around 12%) of participants showed difficulties (Hansson et al., 2007; Sikora & Plapinger, 1994).

Like the literature on vocabulary development, studies examining the morpho-syntactic skills of children with MMHL have also yielded mixed results. On the one hand, studies measuring receptive grammatical abilities using standardised assessments have tended to report no significant group differences between children with MMHL and age-matched

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normally hearing peers (Briscoe et al., 2001; Delage & Tuller, 2007; Gilbertson & Kamhi, 1995; Halliday & Bishop, 2005; Hansson et al., 2004; Moeller et al., 2010; Nittrouer & Burton, 2003; Norbury, Bishop, & Briscoe, 2001, 2002; c.f. Delage & Tuller, 2007). On the other, experimental probes have tended to identify persistent delays and/or deficits in both the perception and production of grammatical morphemes and syntax (Brown, 1984; Delage & Tuller, 2007; DesJardin, Ambrose, Martinez, & Eisenberg, 2009; Hammer & Coene, 2016; Koehlinger, Van Horne, & Moeller, 2013; McGuckian & Henry, 2007; Moeller et al., 2010; Norbury et al., 2001; Tomblin, et al., 2015), including reductions in mean length of utterance (Koehlinger et al., 2013). For instance, such studies have shown that children and/or adolescents with MMHL show delays and/or deviancies in their production of English, French, and Dutch grammatical morphemes (Brown, 1984; Delage & Tuller, 2007; Hammer & Coene, 2016; McGuckian & Henry, 2007; Moeller et al., 2010; Norbury et al., 2001; Tomblin, et al., 2015). Moreover, the limited number of studies that have examined syntactic development in children with mild to severe SNHL have tended to identify patterns of impaired performance even into adolescence (Delage & Tuller, 2007; Elfenbein, Hardin-Jones, & Davis, 1994; Moeller et al., 2010; Tuller & Delage, 2014; Tuller & Jakubowicz, 2004; c.f. Norbury et al., 2002). Whilst individual differences have been underexplored in this literature, several studies have observed that between 30-50% of their MMHL groups showed significant difficulties in standardised measures of morpho-syntactic development (Delage & Tuller, 2007; Koehlinger et al., 2013; Norbury et al., 2001).

Understanding the causes of these mixed results is a complex task. Again, there is little consensus on the role of severity of hearing loss in determining grammatical competence, with some studies finding evidence for a significant effect (Delage & Tuller, 2007; Elfenbein et al., 1994; Koehlinger et al., 2013; Slinger, Grimes, & Christensen, 2010; Wake et al., 2005) and others not (Friedman & Szterman, 2006; Norbury et al., 2001; Tuller

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& Jakubowicz, 2004). Some studies have found a positive influential role of early detection (Kennedy et al., 2006; Sininger et al., 2010; Yoshinaga-Itano et al., 1998), whilst others have not (Norbury et al., 2001; Tuller & Jakubowicz, 2004; Wake et al., 2005). There is also increasing evidence for a positive effect of early intervention (in the form of fitting of hearing aids; Friedman & Szterman, 2006; Koehlinger et al., 2013; Tomblin et al., 2015) as well as hearing aid use (Walker et al., 2015). Compellingly, there now exist a number of studies that have demonstrated delays in the perception and production of specific consonants in children with MMHL, notably, fricatives (McGuckian & Henry, 2007; Moeller et al., 2007; Moeller et al., 2010). These delays have been attributed to limitations in audibility, particularly of female and child talkers, owing to the restricted bandwidth of hearing aids (Moeller et al., 2007; Stelmachowicz, Pittman, Hoover, & Lewis, 2001, 2002). In turn, the resultant inconsistencies in the input have been posited as a potential underlying cause of the difficulties some children with MMHL face in acquiring morphological rules (Moeller et al., 2007; Stelmachowicz, Pittman, Hoover, & Lewis, 2001, 2002). Other researchers have examined the effect of age on grammatical competence in this group. Here, whilst some found that those children who exhibited difficulties in morpho-syntactic development were likely to be younger in age than those who did not (Norbury et al., 2001; Tuller & Jakubowicz, 2004), there is now evidence to suggest that such deficits may in fact persist into adolescence and even adulthood in some individuals (Delage & Tuller, 2007; Huysmans, de Jong, van Lanschot-Wery, Festen, & Goverts, 2014). Finally, some researchers have again speculated that the grammatical difficulties evident in a subset of children with MMHL may be a consequence of a 'double deficit' – the presence of a co-occurring (potentially genetic) risk factor for language impairment that, combined with SNHL, is sufficient to cause significant difficulties (Bishop, 2006; Gilbertson & Kamhi, 1995; Borg, Edquist,



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Reinholdson, Risberg, & McAllister, 2007; Delage & Tuller 2007). However, again, no data exist to verify the legitimacy of this hypothesis.

Lastly, aside from oral language development, the reading and writing skills of children with MMHL have also received some attention in the literature. There are to date a number of studies that have examined the reading and/or phonological skills of children with MMHL. Early studies indicated significant delays and limitations in the literacy outcomes of children with even mild levels of SNHL (Blair, Peterson, & Viehweg, 1985; Davis et al., 1986; Bess, Dodd-Murphy, & Parker, 1998). More recent studies have however tended to yield more positive outcomes, with several studies showing that, on average, children with MMHL obtain reading skills that are either commensurate with their normally hearing peers or with normative means, or both (Briscoe et al., 2001; Gibbs, 2004; Halliday & Bishop, 2005, 2006; Park & Lombardino, 2012, 2013; c.f. Hansson et al., 2004). However, two caveats are worth mention here. First, these studies have typically demonstrated a discrepancy between phonological processing skills and reading, with children with MMHL performing worse than controls on tasks measuring the former (Briscoe et al., 2001; Halliday & Bishop, 2005, 2006; Park & Lombardino, 2012). These studies have suggested that the phonological processing skills of this group worsen with increasing severity of hearing loss (Briscoe et al., 2001; Park & Lombardino, 2012, 2013). Second, there has been some suggestion that different tasks may be differentially affected in this group, with measures of reading rate and rapid naming remaining relatively intact, and reading accuracy and phonological awareness being most affected (Park & Lombardino, 2012, 2013). Further research is needed to verify these claims.

To summarise, the language abilities of children with MMHL are mixed. Studies have shown that as a group, children with MMHL tend to show delays and/or deviancies in their development of phonology, vocabulary, and morpho-syntax, but not in their acquisition of

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reading skills (for reviews, see Delage & Tuller, 2014; Moeller et al., 2007). However, these same children are also characterised by marked individual differences in their performance on these measures. Understanding these individual differences and how to predict them is key to the successful intervention and remediation of these children. The current study aimed to achieve this, by examining a wide range of language abilities in a relatively large sample of children and adolescents with MMHL, and asking which factors were linked to impaired performance. Consistent with the literature, we considered the following factors: age, nonverbal ability, maternal education (as a proxy of socio-economic status; SES), severity and age of confirmation of hearing loss. In addition, to test the hypothesis that children and adolescents with MMHL who go on to show language difficulties might have an additional genetic risk factor, we examined whether family history of language difficulties was a significant predictor (e.g. Carroll, Mundy, & Cunningham, 2014). Specifically, we asked:

1. Do children and adolescents with MMHL have impaired/delayed language relative to (a) their peers, and/or (b) population norms? If so, what aspects of language are affected?
2. What proportion of children and adolescents with MMHL has clinically significant language difficulties? What factors characterise these children?

### **Methods**

#### **Participants**

Two groups of children aged from 8 to 16 years were recruited: children with MMHL (MMHL group) and a control group of typically developing children (CA group). All children were from monolingual English speaking backgrounds and all were required to achieve a minimum T-score of at least 40 on a test of nonverbal ability (i.e. not more than 1 *SD* below the mean, and equivalent to an IQ-score of 85; see below). Ethical approval was

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obtained from the UCL Research Ethics Committee, and informed written consent was obtained from the parent/guardian of each child.

### **MMHL group.**

Fifty-seven children with a diagnosis of bilateral MMHL were recruited for the study. Participants were identified via Peripatetic Services in Local Educational Authorities across London and the South East of England. Information about the study with an invitation to participate was distributed to parents/guardians of children who (a) had a known diagnosis of bilateral MMHL, (b) were aged between 8 and 16 years, (c) were from monolingual English-speaking backgrounds, (d) communicated solely via the oral/aural modality (i.e. did not use sign language), and (e) did not have any other known additional needs. Children whose hearing loss was attributed to neurological impairment were excluded from the study. Those children who met these criteria were invited into UCL for screening. Hearing sensitivity at 250, 500, 1000, 2000, 4000 and 8000 Hz was measured using an Interacoustics AC33 audiometer. Mild hearing loss was defined as a better ear pure-tone average (PTA) threshold of 20-40 dB HL over 250-4000 Hz, and moderate hearing loss as a better ear PTA threshold of 41-70 dB HL (British Society of Audiology, 2011). Note that these criteria typically rule out children who have hearing loss that is confined to the high frequencies (>4 kHz). One child did not meet the criteria for mild or moderate SNHL, and was therefore excluded from the study. A further four did not achieve a nonverbal ability T-score of at least 40 (see below). Six children dropped out of the study prior to completing all testing, and so their data were not included. This left a total sample size for this group of 46 ( $M$  age = 11.44 years,  $SD$  = 2.16; 27 boys, 19 girls; See Table 1). Nineteen of these children had a mild hearing loss ( $M$  = 32.16 dB HL,  $SD$  = 4.84, range = 23 - 40), and 27 had a moderate loss ( $M$  = 51.26 dB HL,  $SD$  = 8.76, range = 41 - 69) according to the guidelines of the British Society of Audiology

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(2011)<sup>1</sup>. Forty-three used a hearing aid in at least one ear. The age of confirmation of SNHL ranged from 2 months to 14 years (median = 57 months;  $M = 54$  months;  $SD = 35.57$ ), and the age of hearing aid fitting from 3 months to 15 years (median = 65 months;  $M = 63$  months;  $SD = 39.60$ ). The late confirmation of hearing loss of some of the children in this study was not surprising because (a) many of the children participating were born prior to the introduction of the UK NHS Newborn Hearing Screening Programme, and (b) even where children were screened, the Programme is designed to detect hearing thresholds  $> 40$  dB HL (i.e. children with mild levels of hearing loss will not be detected). Nevertheless, because the late age of confirmation of some of the children in our study raised the possibility that they had late-onset MMHL, we ran all of the analyses reported here twice: first, including all children, and second, excluding those children whose MMHL was confirmed after 7 years of age ( $n = 6$ ). The results did not change substantially after excluding those children with a late confirmation, and so the only results of the first analysis are reported here.

### **CA group.**

Forty-four control children ( $M$  age = 11.54 years,  $SD = 2.05$ ; 19 boys, 25 girls) with no known hearing loss, educational difficulties, or history of speech and language problems were recruited from primary and secondary schools located in the same geographical locations as those of the children with MMHL (See Table 1). All children had PTA thresholds across octave frequencies 500-4000 Hz of less than 20 dB HL in both ears (British Society of Audiology, 2011), and obtained thresholds no higher than 25 dB HL across each of these frequencies.

### **Procedure**

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<sup>1</sup> Note that PTA thresholds of the MM group would be considered as ranging from slight (16-25 dB) to moderately severe (56-70 dB) according to the American Speech Language Hearing Association (2016).

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Data for the language assessments was collected as part of a larger test battery, for which testing was carried out during two sessions, each lasting approximately 90 minutes, and separated by at least a week. Each child was tested individually by one of two experimenters in a quiet room in the Infant and Child Lab at UCL. In addition, parents/guardians completed two questionnaires about their child's medical, neurological and psychological history. All of the children in the MMHL group who owned a hearing aid used amplification during the psychometric assessments.

### **Questionnaires**

#### **Medical, neurological and psychological history.**

An in-house questionnaire was used to collect information about the medical history of the child (e.g. medical conditions, hearing), any neurological and/or psychological conditions, the language and early development of the child including history of speech and language therapy (SLT), and the child's family history of language and reading problems. A positive family history of speech or language (oral or written) problems was recorded if these were reported in any next-of-kin (a parent or sibling). In addition, age at which the child's mother left full-time education was recorded as a measure of SES.

#### **Communication.**

Communication abilities were assessed using parental report on the Children's Communication Checklist 2<sup>nd</sup> Edition (CCC-2; Bishop, 2003), a parent/teacher checklist designed to screen for communication problems in children aged 4 to 16 years. The checklist consists of 70 items, each comprising a statement of behaviour such as "Leaves off past tense -ed endings". Respondents are asked to judge how often behaviours occur (i.e. less than once a week (or never), at least once a week, once or twice a day, several times a day (or always)). The items reflect behaviours across 10 scales (speech, syntax, semantics, coherence, inappropriate initiation, stereotyped language, use of context, nonverbal communication,

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social relations, interests). Scores on these 10 scales are expressed as scaled scores with a mean of 10 and a standard deviation of 3. Scores on the first eight of these scales are then used to derive a General Communication Composite (GCC), which has been used to identify children likely to have clinically significant language problems; A score of less than 55 on the GCC has been shown to select the bottom 10% of children on communication (Norbury, Nash, Baird, & Bishop, 2004). The discrepancy between scores on the scales of inappropriate initiation, nonverbal communication, social relations, and interests, and the first four scales is used to calculate the Social Interaction Deviance Composite (SIDC). A score of less than 55 on the GCC combined with a negative score on the SIDC has been shown to indicate evidence of an ‘autistic spectrum’ communication profile (Bishop, 2003; Bishop & Norbury, 2002).

### **Psychometric assessments**

#### **Nonverbal ability.**

Nonverbal ability was estimated using the Block Design subtest of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999), which comprises 13 items graded in difficulty. The subtest consists of a set of modelled or pictorial two-dimensional geometric patterns that the participant is required to replicate as quickly as possible within a specified time-limit, using two-colour blocks. Scores are expressed as T-scores with a mean of 50 and a standard deviation of 10.

#### **Phonological memory.**

Phonological processing and short-term memory were assessed using the Repetition of Nonsense Words (*nonword repetition*) subtest from NEPSY (Korkman, Kirk, & Kemp, 1998). For this subtest, 13 nonword items, ranging from two to five syllables in length were presented via a computer at a comfortable listening level. The original items from this subtest were re-recorded by a female native speaker of Southern British English, in a sound

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attenuated booth. The child's task is to repeat each nonword out loud. Because the norms for this test only go up to 12 years 11 months, scores were calculated in two ways. First, raw scores were used to assess whether the MMHL group differed from their peers on this test (*nonword repetition-raw*). Second, we calculated the standard scores ( $M = 10$ ,  $SD = 3$ ) for this test as usual, but used the norms of the oldest age band (12;6 – 12;11) to calculate the standard scores of those children who at the time of testing were aged > 13 years old (MMHL group  $n = 10$ ; CA group  $n = 13$ ; *nonword repetition-SS*). This latter method allowed us to ascertain whether the MMHL group was performing within normal limits for their age, albeit with a restricted upper age limit.

### **Vocabulary.**

Receptive and expressive vocabulary were assessed using the British Picture Vocabulary Scale (BPVS; Dunn, Dunn, Sewell, & Styles, 2009) and the Clinical Evaluation of Language Fundamentals (CELF) subtests Expressive Vocabulary (for children aged 8 – 9 years) and Word Definitions (for children aged 10 upwards) (Semel, Wiig, & Secord, 2006), respectively. In the BPVS, children are presented with an array of four pictures on a test plate, and the experimenter says a word. The child's task is to select the picture that best illustrates the meaning of the word that the experimenter has said. Scores are expressed as standard scores with a mean of 100 and a standard deviation of 15. For the Expressive Vocabulary subtest, children are shown a series of pictures and for each picture the experimenter reads a stimulus phrase (e.g. "what is this?"). The child's task is to say a word that best corresponds to the picture. For each item of the Word Definitions subtest the experimenter says a word and uses it in a sentence. The child's task is to define each of the target words. Participants either completed the Expressive Vocabulary subtest or the Word Definitions subtest, depending on their age. Scores on these tests were termed *expressive*

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*vocabulary* for the purposes of this study. Scores on this variable were expressed as standard scores with a mean of 10 and a standard deviation of 3.

### **Grammar.**

Receptive grammar was assessed using a computerised version of the Test for the Reception of Grammar (TROG-E; Bishop, 2003). The test comprises 20 blocks of four items, with each block measuring understanding of a different grammatical contrast. For each item, comprehension is assessed using a four-item multiple-choice format, where a picture depicting a spoken target sentence is contrasted with foil pictures depicting sentences that are altered in grammatical/lexical structure. The child's task is to select the picture that corresponds to the sentence they have just heard. Scores are expressed as standard scores with a mean of 100 and a standard deviation of 15. Expressive grammar and working memory were assessed using the Recalling Sentences subtest of the CELF (Semel et al., 2006). In this subtest, sentences of increasing length and complexity were presented via a laptop at a comfortable listening level. The child's task is to repeat the sentence verbatim. For this subtest, scores are expressed as standard scores with a mean of 10 and a standard deviation of 3.

### **Reading.**

Word recognition and decoding were assessed using the Word Reading and Pseudoword Decoding subtests of the Wechsler Individual Achievement Test (WIAT; Wechsler, 2005). In these subtests, participants are presented with a series of lists of words or nonwords, and asked to read those words or nonwords out loud as accurately as possible. Scores are expressed as standard scores with a mean of 100 and a standard deviation of 15.

### **Missing data**

It was not possible to obtain a PTA threshold for one child in the CA group owing to poor compliance with the test protocol. Instead, a screening procedure confirmed that this



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child had normal hearing. Questionnaire data recording the age at which the mother left full-time education was missing for five participants (four MMHL, one CA). The CCC-2 was not completed by 10 parents (six CA, four MMHL).

### **Data processing and analysis**

Kolmogorov-Smirnov tests showed that scores were non-normally distributed for the following psychometric assessments: Expressive vocabulary (CA group only), recalling sentences (MMHL group only), receptive grammar (both groups), and pseudoword decoding (CA group only). Parental report scores on the CCC-2 were also not normally distributed. Given that (a) the majority of data sets met the assumptions of normality and (b) parametric statistics are relatively robust to violations of normality, these assessments were analysed using parametric statistics. Data for each test were checked for extreme outliers and none was observed.

## **Results**

### **Group comparisons**

The characteristics of the MMHL and CA groups are presented in Table 1, along with between-group statistical comparisons. As expected, the two groups did not differ on age or maternal education, and the PTA thresholds of the MMHL group were higher (poorer) than those of the CA group. Both groups had a similar number of first-degree relatives with a history of language and/or reading problems, but a greater proportion of the MMHL group had received SLT at some point during their development. However, we also observed that the nonverbal ability scores of the MMHL group, although in the normal range, were significantly lower than those of the CA group. Consequently, where possible (i.e. where parametric statistics were employed), nonverbal ability was used as a covariate in all subsequent group comparisons.

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The parental report scores of the two groups on the ten CCC-2 scales are presented in Table 2, along with between-group statistical comparisons (univariate ANCOVAs controlling for nonverbal ability). For all of the scales, we observed a significant effect of Group after correcting for multiple comparisons (Bonferroni;  $\alpha = .005$ ), with the MMHL group obtaining significantly lower parental report scores than the CA group. This group difference was further reflected in the GCC, with the MMHL group obtaining significantly lower composite scores ( $M = 44.45$ ,  $SD = 21.16$ ) than the CA group ( $M = 81.11$ ,  $SD = 19.44$ ),  $F(1,77) = 51.79$ ,  $p < .001$ ,  $d = .1.89$ , 95% CIs [22.99, 40.58]. Moreover, surprisingly, the mean GCC for the MMHL group was below the 55 cut-off indicating that, on average, the MMHL group fell within the bottom 10% of the general population on parental report of communication skills. In total, of those children with MMHL whose parents completed the CCC-2, 28 (67%) obtained GCC scores that were less than 55. This compared to two (5%) for controls. In contrast, the MMHL group did not differ significantly from the CA group on the SIDC composite measure (MMHL group:  $M = 4.81$ ,  $SD = 7.10$ ; CA group:  $M = 1.18$ ,  $SD = 8.19$ ),  $F(1,77) = 2.25$ ,  $p = .138$ ,  $d = -0.44$ , 95% CIs [-6.14, 0.86]. Nevertheless, we identified five children from the MMHL group (11%) who obtained a GCC score of less than 55, combined with a negative SIDC score, indicating a possible autism spectrum disorder (ASD) profile of communication difficulties. Note that none of the children in this study had a formal diagnosis of ASD.

The scores of the two groups on the seven psychometric language assessments are presented in Table 3. To aid comparison between tests, standard scores are displayed as z-scores ( $M = 0$ ;  $SD = 1$ ) in Figure 1. Three points are evident from the results. First, there was a trend for the MMHL group to obtain lower (poorer) scores than the CA group on all of the seven language assessments tested. To investigate this pattern of results, we ran a series of univariate ANCOVAs with Group (MMHL versus CA) as the between-subjects' variable,

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score as the dependent variable, and nonverbal ability as the covariate (see Table 3). After correcting for multiple comparisons (Bonferroni;  $\alpha = .007$ ), the MMHL group performed significantly more poorly than the CA group on nonword repetition (both the raw and SS measures), expressive vocabulary, receptive grammar, and recalling sentences. Note that these tests all included an aural component. Second, although the MMHL group performed more poorly than the CA group on more than half of the language assessments, they nonetheless on average performed well within normal limits on these tests (i.e. as a group they obtained a mean standard score around the normed mean). Rather, the group differences appeared to be driven by the higher than average performance of the CA group relative to the population mean. The exception here was nonword repetition, where the MMHL group performed significantly below both their peers and the population norms. Finally, it was evident that the MMHL group in general showed greater variability on the language assessments than the CA group. These individual differences will be explored further below, in two ways.

### **Poor performers**

First, we calculated the number of children in each group (MMHL and CA) who obtained standard scores  $> 1 SD$  below the normative mean (corresponding to the bottom 16% of the population) for each of the seven language assessments (so-called *poor performers*). These numbers are displayed in Table 4. A series of Chi-squared analyses showed that the MMHL group had significantly greater proportions of poor performers than the CA group on receptive grammar and nonword repetition. The group difference was particularly evident for nonword repetition, where almost 40% of the MMHL group fell below this cutoff.

We then asked whether any of the children in the MMHL group would meet current criteria for having clinically significant oral or written language difficulties (were it not for

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the presence of a SNHL). To do this, we adopted the criteria set out by McArthur and colleagues (McArthur, Ellis, Atkinson, & Coltheart, 2008) whereby to be classified as having a reading impairment, children had to obtain a standard score that was  $>1 SD$  below the normative mean on at least one of the two reading tests (word reading and pseudoword decoding), but score at least within the average range (standard score  $\leq 1 SD$  below the normative mean) on at least three out of four key spoken language tests (nonword repetition, receptive vocabulary, receptive grammar, recalling sentences). To be classified as having an oral language impairment, children had to obtain a standard score of  $>1 SD$  below the normative mean on at least two of these four key spoken language tests, regardless of their performance on the reading measures. Out of the CA group, two children (5%) met criteria for having a reading impairment and none met the criteria for having an oral language impairment. For the MMHL group, five children (11%) met the criteria for having a reading impairment, and seven (15%) for having an oral language impairment. The proportion of children showing evidence for a reading impairment did not differ significantly between the CA and MMHL groups,  $\chi^2(1) = 1.25, p = .132, OR = 3.09$ . However, the MMHL group contained a significantly higher proportion of children who showed evidence for an oral language impairment than did the CA group,  $\chi^2(1) = 7.26, p = .004, OR = 18.48$ . Note that none of the children in the CA group had received a diagnosis of dyslexia, and none of the children in the MMHL group would have been eligible for a diagnosis of either dyslexia or specific language impairment (SLI) on the basis of them having an SNHL (World Health Organisation, 2015).<sup>2</sup>

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<sup>2</sup> Note that since the initial writing of this manuscript the term *Language Disorder associated with X* has been proposed to include those children whose impairments in language be associated with other conditions (e.g. SNHL); Note that this term is distinct from the term *Developmental Language Disorder* which encompasses those children whose impairments cannot be attributed to a known biomedical aetiology (Bishop, Snowling, Thompson, Greenhalgh, & the CATALISE-2 consortium, 2016).

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Having established that a higher proportion of children with MMHL showed evidence for clinically significant language difficulties than was the case for normally hearing controls, we then asked whether these children differed from their peers in any way. In order to increase power, we combined the reading impairment and oral language impairment subgroups to create a subgroup of children with MMHL who met the criteria for either a reading impairment or an oral language impairment (MMHL-poor;  $n = 12$ ) and a subgroup of children with MMHL who did not (MMHL-normal;  $n = 34$ ). To establish whether the MMHL-poor subgroup did indeed show impaired language (and that the MMHL-normal subgroup did not), we ran a series of univariate ANCOVAs comparing the performance of these two subgroups to that of the CA group on the seven language assessments and on the parental report of communication skills (see Table 5). After controlling for nonverbal ability and adjusting for multiple comparisons (Bonferroni,  $\alpha = .006$ ), there were significant effects of subgroup on all eight language measures. Post-hoc comparisons (LSD) showed that the MMHL-poor subgroup performed significantly more poorly than the CA group on all measures (all  $p$  values  $< .001$ ). The MMHL-poor subgroup also performed significantly more poorly than the MMHL-normal subgroup on the measures of receptive and expressive vocabulary, receptive grammar, recalling sentences, and word and pseudoword reading (all  $p \leq .001$ ). However, the MMHL-poor and MMHL-normal subgroups were not significantly different from each other on the GCC,  $p = .076$ , nor on the test of nonword repetition,  $p = .322$ . Finally, the MMHL-normal subgroup did not differ significantly from the CA group on receptive vocabulary,  $p = .228$ , or on word or pseudoword decoding,  $p = .379$ , and  $p = .856$ , respectively. They did however score more poorly than controls on the GCC,  $p < .001$ , nonword repetition-SS,  $p < .001$ , expressive vocabulary,  $p = .044$ , receptive grammar,  $p = .018$ , and recalling sentences,  $p < .001$ . Note that despite these group differences, the

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MMHL-normal subgroup on average obtained scores that were well within normal limits for all measures apart from the GCC and test of nonword repetition (see Table 5).

We then asked whether any demographic, audiological, or cognitive factors might be linked to the poorer language outcomes of some children with MMHL. To do this, we compared the profiles of the MMHL-poor and MMHL-normal subgroups on a number of variables that have been shown in previous studies to influence language outcomes. These variables were: age, nonverbal ability, mean PTA threshold (i.e. severity of hearing loss), age of confirmation of SNHL, maternal education level, and family history of language and/or reading problems. We also asked whether the two subgroups differed in terms of access to SLT. The results are displayed in Table 6. Three factors distinguished those who showed poor language versus those who showed normal language. First, the MMHL-normal subgroup had better nonverbal ability than the MMHL-poor subgroup. While this difference just missed significance after controlling for multiple comparisons, it nonetheless represented a large effect size of 0.88. Note that as low nonverbal ability was an exclusion factor, all participants had a nonverbal ability score that was within the normal range (i.e. no more than 1 *SD* below the normative population mean). Note also that the mean T-score of the MMHL-poor group was 50 (i.e. equivalent to the normative population mean). The difference therefore appeared to reflect the above average nonverbal ability of the MMHL-normal group as opposed to the below average ability of the MMHL-poor group. Second, there was a trend for an effect of maternal education level, in that the mothers of those children in the MMHL-poor subgroup on average left full-time education slightly earlier than those in the MMHL-normal subgroup. Note that this measure may reflect the slightly higher SES of the MMHL-normal subgroup relative to the MMHL-poor subgroup. Finally, we observed that those children in the MMHL-poor subgroup were more likely to have had a family history of language and/or reading problems than those in the MMHL-normal subgroup. Of those

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children in the MMHL-normal subgroup, only 15% had one or more next-of-kin who had experienced problems with language and/or reading. This figure was 50% for those in the MMHL-poor subgroup.

### Discussion

#### **Mild to moderate sensorineural hearing loss and language ability**

The first aim of the present study was to examine whether children and adolescents with MMHL have impaired/delayed language relative to their peers, and/or population norms. Overall, our results showed that as a group, children and adolescents with MMHL performed significantly more poorly than their normally hearing age-mates on standardised measures of expressive vocabulary, receptive grammar, recalling sentences, and nonword repetition, but not on measures of receptive vocabulary, word or nonword reading. They also scored significantly more poorly than controls on a parent report measure of communication. However, we were careful to include in our test battery assessments that had been recently standardised using UK norms (the exception here was nonword repetition). Close inspection of our data indicated that our MMHL group nonetheless performed on average at or close to the normative mean on the majority of assessments. In fact, it was our control group who performed above average on most of these measures. Nonetheless, for nonword repetition and the parental report of communication abilities (CCC-2), the MMHL group scored both significantly lower than controls and poorer than would be expected for their age based on standardised norms.

Perhaps the most compelling finding of this study was that children and adolescents with MMHL in the main performed *so well* on the standardised language tests. Indeed, on average, the MMHL group scored either at, or marginally below ( $\leq 0.2 SD$ ) the normative mean on standardised measures of receptive and expressive vocabulary and grammar, as well

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as word and nonword reading. This finding is consistent with other relatively recent studies (e.g. Tomblin et al., 2015), and raises the question as to whether or not language difficulties should be considered the norm in children with MMHL. This question has been similarly put forward by Wolgemuh et al. (1998), who argued that the view that language difficulties should be an expected consequence of childhood hearing loss is an erroneous one. Nonetheless, before accepting this conclusion, there are several points of caution that need addressing.

First, our MMHL sample scored on average slightly higher than the norm on both nonverbal ability and SES. Indeed, our MMHL group had an average nonverbal ability T-score that was equivalent to an IQ-score of around 108 (i.e. 0.5 *SD* above the normative mean) and a maternal education level of 19.33 years (UK minimum school leaving age is 16 years). It is therefore likely that poorer outcomes may have been observed had we recruited children from more disadvantaged backgrounds. Indeed, there is some evidence for a cumulative negative effect of low-SES and *temporary* hearing loss (caused by chronic otitis media with effusion) on the language outcomes of children (e.g. Nittrouer, 1996; Nittrouer & Burton, 2005). There is a need for future studies to address the known recruitment bias in the literature in order to examine the effect of MMHL in children and adolescents from low-SES families.

Second, the current study deliberately included older children and adolescents with MMHL, a group that has previously received little attention in the literature (c.f. Delage & Tuller, 2007; Halliday & Bishop, 2005; Sikora & Plapinger, 1994). It may therefore be the case that whilst young children with MMHL show deficits in early language development, they then might 'catch up' with standardised norms either due to developmental effects (e.g. Kiese-Himmel & Reeh, 2006), or following a period of amplification (Koehlinger et al., 2013; Moeller et al., 2010; Tomblin et al., 2015). Indeed, this latter idea gains support from a



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recent large-scale longitudinal study by Tomblin et al. (2015) who investigated the effects of hearing aids on language development in 290 children with mild to severe SNHL. By 6 years of age, the children with SNHL were performing on average around 0.3 *SD* below the normative mean on a battery of standardised measures of language; However, crucially, improved audibility with hearing aids, early fitting of hearing aids, and hearing aid duration in the case of children fitted after 18 months of age were associated with language growth (see also Koehlinger et al., 2013). In the current study, the mean age of intervention was relatively late (63 months), although there were also children who were fitted with hearing aids early (< 18 months) as well as three children who did not wear hearing aids at all. However, because the children in this study were aged between 8 and 16 years old, the vast majority would have experienced a relatively prolonged period of amplification which is likely to have bolstered their language skills.

Third, whilst our MMHL group performed at or slightly below the normative mean on the majority of standardised measures of language we included, they nonetheless performed *above average* on our measure of nonverbal ability. Our MMHL group therefore showed a mismatch between their verbal and nonverbal abilities. One interpretation of this pattern of results is that MMHL may lead to a reduction in language outcomes relative to what might be expected for a normally hearing child in an otherwise identical language-learning environment. This highlights one of the limitations of relying on normative data when drawing conclusions about the outcomes of children with MMHL, and illustrates the importance, where possible, of employing age- and SES matched controls (for similar arguments, see Blair et al., 1985; Tomblin et al., 2015).

In the current study, comparisons between our MMHL group and age-matched controls lead to a very different interpretation of the data than reliance on standardised test norms. Indeed, the mean effect of MMHL on language more than doubled in magnitude (to

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around 0.7 *SD*) when comparing our MMHL group to controls than when comparing it to test norms. This raises the question as to how well-matched our two groups were. It is certainly the case that our control group were unusually unimpaired, with virtually none showing below average performance on any of our psychometric language tasks. However, our control sample had an additional selection criterion that they had no known educational difficulties, or history of speech and language problems, and consequently, they may have been at lower risk of language and reading difficulties than the general population (including our MMHL sample). That said, many studies estimate the incidence of dyslexia or SLI at around 7% in the general population, which is not so different from the 4.5% of poor readers that we saw in our control group (Snowling, 2000; Tomblin, Smith, & Zhang, 1997). It is also the case that our control group performed above average on all of our psychometric measures, a finding which is common in studies such as this one where participants are required to volunteer, and in this case travel to the laboratory for testing. Controls also scored significantly higher than the MMHL group on our measure of nonverbal ability, obtaining an average nonverbal IQ score of 116 (i.e. more than one *SD* above the normative mean). However, this difference was controlled for statistically in our group comparisons. Moreover, although we took care to ensure that our controls were recruited from the same geographical locations as our MMHL group, there was also a trend for our control group to score slightly higher on our measure of SES (with a maternal education level age of 20.5 years). Because this difference was not statistically significant, we did not control for it in the group comparisons; However, doing so did not change the pattern of main effects. It is therefore clear that there were differences between the two groups. However, our findings nonetheless suggest that MMHL in childhood and adolescence may restrict language outcomes, even where group mean scores are within normal limits (Blair et al., 1985; Tomblin et al., 2015).

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Fourth, our results suggest that, amongst other things, measures of phonological processing are likely to pose significant difficulties for children and adolescents with MMHL. Our MMHL group scored around 2 *SD* below controls on our measure of phonological processing, nonword repetition, and 39% obtained standard scores that were > 1 *SD* below the normative mean. These findings are consistent with a number of studies which have demonstrated poorer nonword repetition abilities in children with MMHL relative to both age-matched controls and standardised norms (Briscoe et al., 2001; Halliday & Bishop, 2005, 2006; Park & Lombardino, 2012, 2013). What is surprising however, is how little impact these deficits in nonword repetition appear to have on the general language development of children with MMHL. In the current study, children and adolescents with MMHL showed disproportionate difficulties in nonword repetition relative to the other language measures we included (see also Briscoe et al., 2001). Moreover, when we divided the MMHL group into those who had clinically significant language difficulties versus those who did not, the two subgroups did not differ on nonword repetition.

Our findings contrast against a backdrop of evidence that nonword repetition, and more generally, phonological processing, play a crucial role in both oral and written language development (e.g. Baddeley, Gathercole, & Papagno, 1998). Nonword repetition has been shown to predict oral language outcomes and in particular vocabulary development in both normally hearing children and children with severe to profound SNHL who are fitted with cochlear implants (Baddeley et al., 1998; Casserly & Pisoni, 2013). Moreover, intact phonological representations have been posited as a necessary precursor to learning to read, with deficits in phonological processing being mooted as an underlying cause of developmental dyslexia (see Snowling, 2000; c.f. Boets et al., 2013; Ramus & Szenkovits, 2007). How then might we explain the apparent dissociation between nonword repetition and reading and/or vocabulary acquisition in children with MMHL seen here and elsewhere

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(Briscoe et al., 2001; Halliday & Bishop, 2005, 2006; Park & Lombardino, 2012, 2013)? We can think of three possible explanations for this. First, it may be that the phonological working memories and/or representations of children with MMHL are intact, but that nonword repetition is not sufficiently sensitive to gauge this. Indeed, nonword repetition requires the execution of a number of complex skills, including the rapid encoding, decomposition, manipulation, and articulation of linguistic units, and a breakdown at any of these points will lead to diminished performance. Given its reliance on auditory input, it is possible that nonword repetition underestimates the phonological skills of children with MMHL, and indeed it is notable that those studies that have included tasks that rely more on the visual modality (e.g. rapid naming) have failed to find a difference between MMHL and control groups on these measures (Halliday & Bishop, 2006; Park & Lombardino, 2012). Second, it may be that theories have over-stated the importance of intact phonological representations and short-term memory acquisition in oral and written language development, in that children can display deficits in phonological processing in the absence of other major deficits (e.g. Mody et al., 1999; Stothard, Snowling, & Hulme, 1996). Finally, it is possible that for both oral and written language development, deficits only emerge as a result of an interaction between a number of risk and protective factors (Bishop, 2006; Pennington, 2006). We consider this latter possibility below.

Aside from phonological processing, our study also found that 67% of children and adolescents with MMHL had significant communication problems as measured by parental report. One interpretation of these findings is that parents of children and adolescents with MMHL are more likely to over-report negative communication behaviours, because of pre-existing expectations about the likely impact of MMHL on their child's language abilities. However, there is now increasing evidence that standardised laboratory measures of language and literacy may be functionally distinct from more 'real-world' measures (DeThorne et al.,

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2008; Tomblin et al., 2015). There is now a handful of studies that have reported deficits in the morpho-syntactic abilities of children with MMHL who nonetheless perform largely within the normal range on standardised measures of expressive and/or receptive grammar (Koehlinger et al., 2013; Moeller et al., 2010; Tomblin, et al., 2015). Moreover, studies examining the spontaneous speech of children and adults with MMHL have shown evidence for a reduction in syntactic complexity and an increase in morphological errors (Huysmans et al., 2014; Koehlinger et al., 2013; Tomblin et al., 2015). It may be therefore that standardised laboratory-based language assessments fail to capture the extent of difficulties that children and adolescents have with communication in every-day life.

Finally, our results showed that whilst on average children with MMHL performed within normal limits on the majority of measures of language and literacy, there was nonetheless a significant proportion who did not. Our findings add to an increasing body of evidence that whilst children with MMHL may not *necessarily* show deficits in language development, they are nonetheless at *greater risk* of experiencing difficulties with language than their normally hearing peers. We consider this proposal below.

### **Poor performers**

Having established that poor language outcomes were not the norm in children with MMHL, the second aim of the present study was to examine the proportion of children and adolescents with MMHL that had clinically significant language difficulties, and to ask what factors characterised these children? We found evidence for substantial individual differences in performance amongst the MMHL group, with some children performing well below expected levels, and others within or even above normal limits. When we identified poor performers as a function of language task, we found that around between 9-17% of the MMHL group performed more than 1 *SD* below the normative mean on any given task (the exception here was nonword repetition, where 39% performed below this cutoff). When we

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identified participants who performed poorly on more than one standardised assessment, we observed that 26% of our MMHL group respectively showed a profile of performance that was suggestive of having either a clinically significant reading or oral language deficit. Our estimates are therefore on par with those studies which have identified the proportions of poor performers in children with MMHL as anywhere between 12-60% (Briscoe et al., 2001; Delage & Tuller, 2007; Gilberton & Kamhi, 1995; Kiese-Himmel & Reeh, 2006; Norbury et al., 2001; Sikora & Plapinger, 1994).

In terms of factors characterising these poor performers, we found that children with MMHL who showed clinically significant language problems were more likely to have a family history of language and/or reading problems that was unrelated to hearing than those who did not. There were also non-significant (albeit medium to large sized) effects for poor performers to have lower nonverbal ability and mothers who left school at a younger age relative to children with MMHL who showed normal/good language skills. The influence of maternal education was not entirely surprising. Indeed, there is mixed evidence for the role of parent education in influencing language outcomes in children with SNHL, with some studies finding an effect (Fitzpatrick, Durieux-Smith, Eriks-Brophy, Olds, & Gaines 2007; Geers, 2002) and others not (Mayne et al., 1998a, 1998b). It is possible that these inconsistencies have arisen because parent/maternal education levels do not have a direct effect on language outcomes, but rather represent a difference in access to services, language-input, or in parent-child interaction. For instance, there is some evidence that greater family participation in children's early intervention programs may mediate the relationship between maternal education and language (Sarant, Holt, Dowell, Rickards, & Blamey, 2009).

Regarding the role of nonverbal ability, there is now a body of evidence that nonverbal ability is a strong predictor of language outcomes in children with hearing loss (Geers, 2002; Mayne et al., 1998a, 1998b; Sarant, Hughes & Blamey, 2010; Sarant et al.,

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2009). However, our results may further our understanding of this relationship, given the restricted range of abilities that we included; All children in this study had a nonverbal ability score of at least 85 and, consequently, the poorer performance of the MMHL-poor subgroup cannot be explained simply in terms of them having low IQ (indeed, the mean nonverbal ability score for this subgroup was precisely at the population mean of 100). By way of contrast, the MMHL-normal subgroup had a mean nonverbal ability score equivalent to 112. It may therefore be that *high* nonverbal ability supports the language development of children and adolescents with MMHL rather than that low nonverbal ability inhibits it. According to this model we may interpret high nonverbal ability as comprising a *protective factor* in the development of normal language abilities in children with MMHL.

Finally, we also set out to establish whether the poor language outcomes of some children with MMHL may be the result of an additional genetic risk factor which co-occurs alongside a child's SNHL. This argument has been put forward by several researchers (e.g. Gilbertson & Kamhi, 1995; Borg et al. 2007; Delage & Tuller 2007) and yet, to date, there has been no evidence in support of this claim. Our finding that 50% of the MMHL-poor group had a first degree relative with a history of language or reading problems (compared to just 15% for the MMHL-normal group) provides just this. However, in addition, our data suggest that the language impairments experienced by these children are unlikely to be independent from their SNHL, because the proportions of children affected are higher than we would expect for the general (normally hearing) population. Children with MMHL are just as likely as those with normal hearing to inherit one or more genetic risk factors for the development of an oral or written language impairment (indeed, there was no difference in family history of language impairments between our MMHL and CA groups). However, if they do, our findings suggest that these risk factors are likely to interact with the impoverished language environment available to children with MMHL as a result of their

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hearing loss, leading to a greater likelihood that the child will go on to experience poor language outcomes. This explanation is consistent with two multiple risk factor models for oral and written language impairments respectively, that have been put forward in the literature (Bishop, 2006; Pennington, 2006). Our findings further contribute to these models by providing preliminary evidence that whereas MMHL, family history of language problems and lower SES might constitute risk factors for the development of clinically significant language problems in children, better nonverbal ability might constitute a protective factor.

### **Conclusion**

As a group, children with MMHL on average performed more poorly than their normally hearing peers but nonetheless generally within normal limits on a range of language tasks. The exceptions were nonword repetition and parental report of communication abilities, for which children with MMHL performed worse than both controls and population norms. However, we saw evidence of considerable individual differences, with some children with MMHL performing within normal limits and others showing evidence of clinically significant oral or written language difficulties. Of the variables we investigated, nonverbal ability, maternal education, and family history of language problems appeared to be linked to language difficulties in children with MMHL. Our results suggest that whether or not an individual child with MMHL goes on to develop language difficulties is likely to be determined by the interaction between a number of risk and protective factors. Predicting those children that are at increased risk of going on to have clinically significant language difficulties is key to the future clinical management of this group.

### **Acknowledgements**

The project work was funded by an ESRC First Grants Award (RES-061-25-0440). Thanks to Steve Nevard and Kathleen McCarthy for their assistance in recording some of the stimuli.



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Thanks also to Páraic Scanlon for testing some of the participants. Special thanks to all of the children who participated, along with their parents, as well as the Local Educational Authorities and schools who assisted with recruitment.

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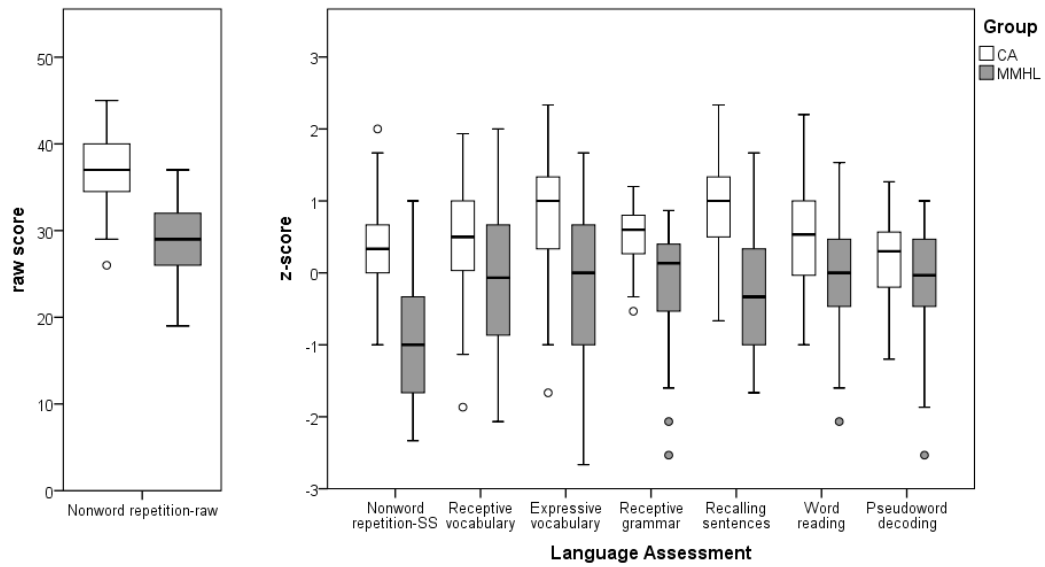
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**Figure Caption**

Figure 1: Boxplots showing scores on the psychometric language assessments for the CA group (white) and MMHL group (grey). The left-hand plot displays raw scores on the nonword repetition task. For ease of comparison between tests, standard scores are displayed as z-scores ( $M = 0$ ,  $SD = 1$ ) in the right hand plot. The black line inside each box denotes the 50<sup>th</sup> percentile, and the lower and upper box boundaries the 25<sup>th</sup> and 75<sup>th</sup> percentiles respectively of each distribution. The whiskers above and below the box boundaries indicate the largest and smallest observed values that are not statistical outliers.

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## LANGUAGE DEVELOPMENT IN CHILDREN WITH HEARING LOSS

Table 1: Participant characteristics and between-group comparisons

| Variable                              | CA (n = 44) |           | MMHL (n = 46) |           | Statistic (df)      | <i>p</i>        | Effect size | 95% CI           |
|---------------------------------------|-------------|-----------|---------------|-----------|---------------------|-----------------|-------------|------------------|
|                                       | <i>M</i>    | <i>SD</i> | <i>M</i>      | <i>SD</i> |                     |                 |             |                  |
| Age (years)                           | 11.54       | 2.05      | 11.44         | 2.16      | $t(88) = 0.23$      | .821            | 0.05        | [-0.78, 0.98]    |
| PTA threshold (dB) <sup>a</sup>       | 8.85        | 4.13      | 46.00         | 11.92     | $t(56.30) = -19.89$ | <b>&lt;.001</b> | -8.99       | [-40.88, -33.40] |
| Maternal education (age) <sup>b</sup> | 20.47       | 2.89      | 19.33         | 2.65      | $t(83) = 1.88$      | .063            | 0.39        | [-0.65, 2.33]    |
| Nonverbal ability (SS)                | 60.64       | 8.48      | 55.63         | 8.71      | $t(88) = 2.76$      | <b>.007</b>     | 0.59        | [1.40, 8.61]     |
| SLT (n)                               | 10          |           | 31            |           | $\chi^2(1) = 18.09$ | <b>&lt;.001</b> | 7.03        | [2.75, 17.93]    |
| Family history (n)                    | 9           |           | 11            |           | $\chi^2(1) = 0.16$  | .347            | 1.22        | [0.45, 3.32]     |

*Note.* CA = children with typical development (control); MMHL = mild to moderate sensorineural hearing loss group; effect size = Cohen's *d* for *t*-tests, and odds ratio (OR) for chi-squared tests; CI = confidence interval; age = *M* of session 1 and 2; PTA = pure tone average (*M* across left and right ears); maternal education = age (years) at which mothers left full-time education; nonverbal ability was assessed using the Block Design subtest of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999; see text); SS = standard score; SLT = Speech and language therapy (either discontinued or ongoing); family history = children with a next-of-kin (parent or sibling) with oral language and/or reading problems. All comparisons on scale data (age, PTA threshold, maternal education, nonverbal ability) were *t*-tests. Group comparisons on SLT and family history were done using Chi-squared tests (one-sided). All significant comparisons ( $p < .05$ ) remained so after controlling for multiple comparisons (Bonferroni;  $\alpha = .008$ ; boldface).

<sup>a</sup>CA group n = 43; <sup>b</sup>MMHL group n = 42 and CA group n = 43.

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Table 2: Mean parent ratings and between-group comparisons on the CCC-2 scales

| Scale                    | CA (n = 38) |           | MMHL (n = 40) |           | <i>F</i> (1) | <i>p</i>        | Cohen's <i>d</i> | 95% CI       |
|--------------------------|-------------|-----------|---------------|-----------|--------------|-----------------|------------------|--------------|
|                          | <i>M</i>    | <i>SD</i> | <i>M</i>      | <i>SD</i> |              |                 |                  |              |
| Speech                   | 8.30        | 3.34      | 3.76          | 2.53      | 37.35        | <b>&lt;.001</b> | 1.36             | [2.62, 5.15] |
| Syntax                   | 9.98        | 2.88      | 5.00          | 2.72      | 54.54        | <b>&lt;.001</b> | 1.73             | [3.27, 5.68] |
| Semantics                | 11.30       | 3.24      | 6.39          | 4.44      | 25.10        | <b>&lt;.001</b> | 1.52             | [2.60, 6.03] |
| Coherence                | 9.05        | 3.19      | 5.41          | 3.29      | 18.72        | <b>&lt;.001</b> | 1.14             | [1.67, 4.51] |
| Inappropriate initiation | 11.28       | 3.01      | 7.20          | 3.57      | 24.42        | <b>&lt;.001</b> | 1.36             | [2.21, 5.19] |
| Stereotyped language     | 10.80       | 3.03      | 7.20          | 3.76      | 16.31        | <b>&lt;.001</b> | 1.19             | [0.54, 4.52] |
| Use of context           | 10.05       | 3.30      | 3.86          | 2.75      | 75.26        | <b>&lt;.001</b> | 1.88             | [4.25, 6.78] |
| Nonverbal communication  | 10.05       | 2.92      | 5.77          | 2.64      | 39.42        | <b>&lt;.001</b> | 1.47             | [2.56, 4.93] |
| Social                   | 9.10        | 3.32      | 5.81          | 3.71      | 14.12        | <b>&lt;.001</b> | 0.99             | [1.41, 4.59] |
| Interests                | 9.60        | 3.13      | 6.43          | 3.04      | 18.63        | <b>&lt;.001</b> | 1.01             | [1.63, 4.43] |

*Note.* CA = children with typical development (control); MMHL = mild to moderate sensorineural hearing loss group; CI = confidence interval. All comparisons were univariate ANCOVAs controlling for nonverbal ability, and all contrasts remained significant after controlling for multiple comparisons (Bonferroni;  $\alpha = .005$ ; boldface).



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Table 3: Mean scores and between-group comparisons on the psychometric language assessments

| Variable               | CA (n = 44) |       | MMHL (n = 46) |       | F(1)  | p               | Cohen's d | 95% CI        |
|------------------------|-------------|-------|---------------|-------|-------|-----------------|-----------|---------------|
|                        | M           | SD    | M             | SD    |       |                 |           |               |
| Nonword repetition-raw | 36.75       | 4.22  | 28.85         | 4.67  | 58.71 | <b>&lt;.001</b> | 1.87      | [5.49, 9.33]  |
| Nonword repetition-SS  | 11.11       | 1.82  | 7.35          | 2.55  | 52.57 | <b>&lt;.001</b> | 2.52      | [2.52, 4.42]  |
| Receptive vocabulary   | 107.98      | 11.99 | 98.15         | 15.12 | 5.44  | .022            | 0.82      | [0.94, 11.75] |
| Expressive vocabulary  | 12.48       | 2.56  | 9.76          | 3.14  | 12.10 | <b>.001</b>     | 1.06      | [0.85, 3.12]  |
| Receptive grammar      | 107.64      | 6.71  | 98.72         | 11.99 | 12.08 | <b>.001</b>     | 1.33      | [3.04, 11.20] |
| Recalling sentences    | 12.80       | 1.91  | 9.28          | 2.37  | 47.53 | <b>&lt;.001</b> | 1.84      | [2.25, 4.07]  |
| Word reading           | 106.68      | 11.05 | 99.43         | 11.19 | 5.33  | .023            | 0.66      | [0.77, 10.17] |
| Pseudoword decoding    | 102.91      | 9.09  | 97.46         | 11.96 | 2.19  | .143            | 0.60      | [-1.12, 7.63] |

*Note.* CA = children with typical development (control); MMHL = mild to moderate sensorineural hearing loss group; CI = confidence interval; SS = standard score. All comparisons were univariate ANCOVAs controlling for nonverbal ability. Comparisons that remained significant after controlling for multiple comparisons (Bonferroni;  $\alpha = .006$ ) are in boldface. All scores are standard scores except for nonword repetition-raw.

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Table 4: Number of participants and group comparisons of poor performers on the psychometric language assessments

| Variable              | CA (n = 44) | MMHL (n = 46) | $\chi^2(1)$ | <i>p</i>         | Odds ratio |
|-----------------------|-------------|---------------|-------------|------------------|------------|
| Nonword repetition-SS | 0           | 18            | 21.52       | <b>&lt; .001</b> | 18.50      |
| Receptive vocabulary  | 2           | 8             | 3.76        | .027             | 4.42       |
| Expressive vocabulary | 1           | 7             | 4.65        | .016             | 7.72       |
| Receptive grammar     | 0           | 7             | 7.26        | <b>.004</b>      | 16.90      |
| Recalling sentences   | 0           | 4             | 4.00        | .022             | 9.42       |
| Word reading          | 0           | 5             | 5.06        | .012             | 11.80      |
| Pseudoword decoding   | 2           | 7             | 2.85        | .046             | 3.77       |

*Note.* CA = children with typical development (control); MMHL = mild to moderate sensorineural hearing loss group; SS = standard score. All comparisons were one-sided chi-squared. Comparisons that remained significant after controlling for multiple comparisons (Bonferroni;  $\alpha = .007$ ) are shown in boldface.

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Table 5: Mean scores and between-subgroups comparisons on the language assessments

| Variable                           | CA (n = 44) |           | MMHL-normal (n = 34) |           | MMHL-poor (n = 12) |           | <i>F</i> | <i>p</i>        |
|------------------------------------|-------------|-----------|----------------------|-----------|--------------------|-----------|----------|-----------------|
|                                    | <i>M</i>    | <i>SD</i> | <i>M</i>             | <i>SD</i> | <i>M</i>           | <i>SD</i> |          |                 |
| GCC <sup>a</sup>                   | 81.11       | 19.44     | 49.39                | 19.60     | 30.55              | 19.87     | 28.26    | <b>&lt;.001</b> |
| Nonword repetition <sup>a</sup>    | 11.11       | 1.82      | 7.65                 | 2.58      | 6.50               | 2.36      | 26.78    | <b>&lt;.001</b> |
| Receptive vocabulary <sup>b</sup>  | 107.98      | 11.99     | 103.06               | 13.27     | 84.25              | 11.03     | 9.71     | <b>&lt;.001</b> |
| Expressive vocabulary <sup>c</sup> | 12.48       | 2.56      | 11.12                | 2.21      | 5.92               | 1.98      | 24.65    | <b>&lt;.001</b> |
| Receptive grammar <sup>c</sup>     | 107.64      | 6.71      | 101.97               | 8.77      | 89.50              | 15.23     | 12.74    | <b>&lt;.001</b> |
| Recalling sentences <sup>c</sup>   | 12.80       | 1.91      | 10.00                | 2.30      | 7.25               | 1.06      | 33.00    | <b>&lt;.001</b> |
| Word reading <sup>b</sup>          | 106.68      | 11.05     | 104.18               | 7.51      | 86.00              | 8.76      | 16.26    | <b>&lt;.001</b> |
| Pseudoword decoding <sup>b</sup>   | 102.91      | 9.09      | 102.47               | 7.20      | 83.25              | 11.45     | 18.16    | <b>&lt;.001</b> |

*Note.* CA = children with typical development (control); MMHL = mild to moderate sensorineural hearing loss group; GCC = General Communication Composite. All comparisons were univariate ANCOVAs controlling for nonverbal ability, and all were done using standard scores. All comparisons remained significant after controlling for multiple comparisons (Bonferroni;  $\alpha = .006$ ; boldface).

<sup>a</sup>CA > MMHL-normal = MMHL-poor. <sup>b</sup>CA = MMHL-normal > MMHL-poor; <sup>c</sup>CA > MMHL-normal > MMHL-poor (Least Significant Difference;  $p < .05$ ).

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Table 6: participant and audiological characteristics and between-subgroups comparisons

| Variable                 | MMHL-normal (n = 34) |           | MMHL-poor (n = 12) |           | Statistic (df)     | <i>p</i>    | Effect size | 95% CI          |
|--------------------------|----------------------|-----------|--------------------|-----------|--------------------|-------------|-------------|-----------------|
|                          | <i>M</i>             | <i>SD</i> | <i>M</i>           | <i>SD</i> |                    |             |             |                 |
| Age (years)              | 11.44                | 2.13      | 11.44              | 2.36      | $t(44) = 0.00$     | .998        | 0.00        | [-1.48, 1.48]   |
| PTA threshold (dB)       | 47.10                | 11.87     | 42.83              | 12.01     | $t(44) = 1.07$     | .291        | 0.36        | [-3.79, 12.33]  |
| Age diagnosis (months)   | 50.67                | 32.91     | 64.58              | 41.89     | $t(43) = -1.17$    | .250        | -0.42       | [-38.00, 10.17] |
| Maternal education (age) | 19.87                | 2.66      | 17.82              | 2.04      | $t(40) = 2.33$     | .025        | 0.77        | [0.27, -3.84]   |
| Nonverbal ability (SS)   | 57.59                | 8.50      | 50.08              | 6.96      | $t(44) = 2.75$     | .009        | 0.88        | [2.00, 13.01]   |
| SLT (n)                  | 23                   |           | 8                  |           | $\chi^2(1) = 0.00$ | .475        | 0.96        | [0.24, 3.87]    |
| Family history (n)       | 5                    |           | 6                  |           | $\chi^2(1) = 6.07$ | <b>.007</b> | 5.80        | [1.32, 25.40]   |

*Note.* All comparisons on scale data (age, PTA threshold, maternal education, nonverbal ability) were *t*-tests. Subgroup comparisons on proportion data (SLT and family history) were done using Chi-squared tests (one-sided). Comparisons that remained significant after controlling for multiple comparisons (Bonferroni;  $\alpha = .007$ ) are in boldface. Age = *M* of session 1 and 2; PTA = pure tone average (*M* across left and right ears); Age diagnosis = age (months) at which SNHL was diagnosed; Maternal education = age (years) at which mothers left full-time education; SS = standard score; SLT = speech and language therapy (either discontinued or ongoing); Family history = children with a next-of-kin (parent or sibling) with oral language and/or reading problems. Effect size = Cohen's *d* for *t*-tests, and OR for Chi-squared tests. CI = confidence interval.