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Mild-Moderate Congenital Hearing Loss: Secular Trends in Outcomes Across Four Systems of Detection

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Abbreviations: UNHS, Universal Newborn Hearing Screening; dB PTA, decibels pure tone average; dB HL, decibels hearing level; CI, confidence interval

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Contributors' Statement:

Mr Carew conceptualized and designed the study, was responsible for the acquisition and interpretation of data, drafted and revised the manuscript.

Dr Mensah provided guidance with analysis and interpretation of the data, and reviewed and revised the manuscript.

Associate Professor Rance assisted with the design of the study, assisted with interpretation of the data and reviewed and revised the manuscript.

Drs Flynn and Poulakis provided guidance regarding data collection instruments and reviewed and revised the manuscript.

Professor Wake led the CHIVOS, SCOUT and VicCHILD cohorts, conceptualized the study, provided guidance regarding data collection instruments, analysis and interpretation of the data and reviewed and revised the manuscript.

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ABSTRACT

Background: Universal newborn hearing screening (UNHS) targets moderate or greater hearing loss. However, UNHS also frequently detects children with mild loss that results in many receiving early treatment. The benefits of this approach are not yet established. We aimed to (1) compare language and psychosocial outcomes between four hearing loss detection systems for children aged 5-8 years with congenital mild-moderate hearing loss; (2) determine whether age of detection predicts outcomes; and (3) compare outcomes between children identified via well-established UNHS and the general population.

Methods: Linear regression adjusted for potential confounding factors was used throughout. Via a quasi-experimental design, language and psychosocial outcomes were compared across four population-based Australian systems of hearing loss detection: opportunistic detection, born 1991-1993, n=50; universal risk factor referral, born 2003-2005, n=34; newly established UNHS, born 2003-2005, n=41; and well-established UNHS, born 2007-2010, n=21. In pooled analyses, we examined whether age of detection predicted outcomes. Outcomes were similarly compared between the current well-established UNHS system and typically-developing children in the Early Language in Victoria Study, born 2003, n=1217.

Results: Age at diagnosis and hearing aid fitting fell steadily across the four systems. For moderate losses, mean expressive language (p for trend .05) and receptive vocabulary (p for trend .06) improved across the four systems, but benefit was not obvious for mild losses. In pooled analyses, diagnosis before age six months predicted better language outcomes for moderate losses. Children with mild-moderate losses exposed to well-established UNHS

continue to experience expressive language scores well below children in the general population (adjusted mean difference -8.9 points, 95% CI -14.7 to -3.1).

Conclusions: Treatment arising from UNHS appears to be clearly benefitting children with moderate hearing losses. However, rigorous trials are needed to quantify benefits, versus costs and potential harms, of early aiding of children with mild losses.

INTRODUCTION

Early diagnosis and amplification offers the potential to prevent, rather than remediate, language disability resulting from bilateral congenital hearing loss (Wake and Carew 2016). Universal newborn hearing screening (UNHS) has now revolutionized the management landscape across the entire spectrum of hearing loss. This includes children born with mild and moderate loss, whose language outcomes show some evidence of better performance when identified early, but on average remain poorer than population norms. For example, in the contemporaneous Outcomes of Children with Hearing Loss (OCHL) study, children with mild and moderate loss had outcomes on average between two-thirds and one standard deviation below their normally hearing peers at age 5 years (Tomblin *et al.* 2015). Therefore it is not surprising that audiologic practice has rapidly encompassed amplification for milder losses that were not initially the target condition for UNHS (Bagatto *et al.* 2010; King 2010).

Any treatment, including early fitting of amplification, should only become routine practice when there is clear evidence that benefits are cost-effective and exceed any treatment burden, stress or negative impact on quality of life (Grimes and Schulz 2002). There is evidence from quasi-randomized and observational studies of better language outcomes when intervention for hearing loss occurs early (Nelson *et al.* 2008), but also that these benefits are greater with more severe losses (Ching 2015). This suggests the maximal benefit of very early detection and amplification may not be as sizeable for mild losses, when compared to the benefit expected for moderate losses. Quantifying this potential benefit requires targeted research at the milder end of the hearing loss spectrum. Unfortunately,

these children comprise a historically less-researched group than those with severe and profound loss (Moeller *et al.* 2015; Stika *et al.* 2015).

Ideally, this evidence would be attained by conducting randomized experimental trials of amplification versus no amplification, stratified by degree of loss. However, the surging number of hearing aids fitted for very young children with a mild loss in the better hearing ear has occurred in the absence of such trials (Australian Hearing, 2015). At this point, therefore, the best evidence may come from natural experiments, i.e. quasi-experimental studies that compare outcomes of different detection systems within populations that are otherwise similar. Observational longitudinal studies can also provide valuable evidence regarding age of amplification, although there is substantial potential for residual confounding which is likely to promote over-estimates of efficacy (Craig *et al.* 2012).

Four Australian population-based state-wide detection systems provide the opportunity to compare both quasi-experimental and longitudinal approaches to the question, "Does earlier diagnosis and amplification benefit children with mild and moderate hearing loss?". It is also important to know the extent of deficits these children continue to experience. Over two decades, these detection systems advanced from opportunistic detection, to risk factor screening, through to 'newly established' UNHS and to the current 'mature' (well-established) UNHS system. Therefore, for four population-derived samples of 5 to 8 year old children with mild or moderate bilateral hearing loss believed congenital, we aimed to:

1. Compare mean language, behaviour and quality of life outcomes between detection systems,
2. Determine whether age of detection predicts these outcomes in the pooled sample, and
3. Compare mean outcomes between the sample exposed to the mature UNHS program and a contemporaneous, typically developing cohort.

METHODS

This was primarily a quasi-experimental study utilizing state-wide population cohorts of children with suspected congenital hearing loss. Cohorts were separated by year and/or geographic state of birth. Victoria's Royal Children's Hospital Ethics Committee approved all studies, and parents provided written informed consent.

Population samples with hearing loss: Over nearly 20 years, Wake et al have recruited four distinct cohorts capturing the evolution in detection systems in two Australian states that mirror international changes. The states share similarities in birth cohort size (approximately 75,000 and 88,000 annually in Victoria and New South Wales (NSW) respectively), socio-demographic characteristics and access to diagnostic and intervention services (Wake *et al.* 2016).

To assemble the cohorts, researchers approached parents of all children registered in state-wide databases as having mild-profound congenital hearing loss that was believed to be

present from birth or soon after. The first three cohorts are historical, while the fourth reflects the current UNHS program in Victoria:

1. Opportunistic - Victorian children born January 1991-July 1993, in the Children with Hearing Impairment in Victoria Outcome Study (CHIVOS) (Wake *et al.* 2004; Wake *et al.* 2005). No effective systematic approach to detecting congenital hearing loss existed.
2. Risk Factor - Victorian children born March 2003-February 2005, in the Statewide Comparison of Outcomes (SCOUT) quasi-randomized trial (Wake *et al.* 2016). Victoria offered automated auditory brainstem response (AABR) screening only for newborns admitted to the neonatal intensive care unit, complemented by risk factor screening at ages 2 weeks and 8 months, and behavioural hearing screening at age 8-10 months.
3. Newly Established UNHS - SCOUT participants born March 2003-February 2005 in NSW, immediately after state-wide UNHS implementation with AABR screening.
4. Mature UNHS - children born April 2007-April 2010 and participating in the state-wide Victorian Childhood Hearing Impairment Longitudinal Databank (VicCHILD), open to all children born with hearing loss in Victoria. The UNHS program was similar to that of Group 3, but with greatly streamlined follow-up processes that minimise delays between identification and accessing services.

These four cohorts were subsequently pooled to conduct longitudinal analyses for our second aim.

Population sample without hearing loss: The Typically Developing group comprised participants from the Early Language in Victoria Study (ELVS) (Reilly *et al.* 2006), born and recruited in 2003 from six metropolitan local government areas. Australian census data indicated that these spanned the disadvantage-advantage spectrum, thus maximising population representation. The inclusion of this cohort allowed analyses addressing our third aim.

All source cohorts excluded families with insufficient English to participate (e.g. questionnaires written at Grade 6 level). CHIVOS additionally excluded children with known intellectual disability. Of the hearing loss cohorts, only VicCHILD is open to children who have never used a hearing device. SCOUT and CHIVOS required fitting by 4 and 4.5 years of age respectively.

Sampling and Procedures: Participants were aged between 5 and 8 years at assessment, and all (except the typically developing group) were classified as having either mild (20 to 40 decibels pure tone average (dB PTA)) or moderate (41 to 65 dB PTA) hearing loss in the better ear at birth or from soon after. Researchers audited individual study databases for children who met inclusion and exclusion criteria, extracting assessment data for the Opportunistic, Risk Factor, Newly Established UNHS and Typically Developing groups. Procedures to obtain these data were consistent with those outlined for the Mature UNHS group below.

For the Mature UNHS group (VicCHILD sourced), eligible families were approached for participation at which point the expected time commitment was outlined (i.e. a two-hour home visit). Families either agreed to fully participate or opted to complete parent questionnaires only. Children used their usual amplification during assessment if applicable.

Measures: Outcome measures are shown in Table 1. At age 5 to 7 years, Mature UNHS participants were directly assessed on standardized measures of language, receptive vocabulary, and non-verbal IQ. Parents reported their child's behaviour and health-related quality of life. Newly Established UNHS/Risk Factor and Opportunistic Detection participants were similarly assessed in language, receptive vocabulary and IQ at age 5 to 6 years and 7 to 8 years respectively. Data from Typically Developing participants at 7 years of age were also accessed for similar domains. Children from the risk factor, newly established and mature UNHS groups whose physical or intellectual disabilities precluded direct assessment were assigned the following basal scores: receptive/expressive language (basal score=50; n=13, 9%), receptive vocabulary (basal score=20; n=9, 6%).

Potential *a priori* confounders covered demographic factors of child sex, parent education level, English as the child's second language and the Australian census-based Disadvantage Index (SEIFA, national mean 1000, SD 100; higher scores reflect less disadvantage) (Australian Bureau of Statistics 2008). Others were non-verbal IQ (intellectual disability score <70 or unable to be assessed) and current hearing loss (most recent better ear three-frequency PTA).

Statistical Analysis: For Aim 1 we estimated mean group differences between all outcomes using linear regression, adjusting for demographic and then the non-demographic potential confounders listed above. We conducted analyses with all children from the Mature UNHS, Newly Established UNHS and Risk Factor groups, and then repeated analyses only for children without intellectual disability. This facilitated inclusion of the Opportunistic group, as this cohort originally excluded children with intellectual disability. Plotting fully adjusted means with 95% confidence intervals enabled visualization of trends across the systems of detection against reference means, accompanying tests of significance are also presented.

Aim 2 drew on pooled data from the four hearing loss groups. Fractional polynomials graphically and empirically examined whether age at detection may influence outcomes, independent of detection system, among children without intellectual disability. Linear regression with identical adjustment to Aim 1 quantified the extent to which continuous outcomes were predicted by age of detection, categorised ordinally into a 0-6 month period and then in 12 month divisions to achieve even participant distributions.

For Aim 3, adjusted linear regression (as above) estimated mean differences in language outcomes between the Mature UNHS and Typically Developing groups.

RESULTS

Figure 1 shows the participant flow for the four groups. Detailed recruitment figures for the three historical groups are reported elsewhere (Wake *et al.* 2004; Wake *et al.* 2016); from the 84 children aged 5-7 years registered with VicCHILD, 31 had mild/moderate hearing loss of whom 67% participated in the Mature UNHS group.

Initial and current hearing losses and non-verbal IQ were similar across the groups (Table 2). A total of 15 children (10.3%) were categorized with intellectual disability: 2 (9.5%) in the Mature UNHS and 13 (17.3%) in the Newly Established UNHS and Risk Factor groups. All children in the historical groups and 19 of the 20 in the Mature UNHS group used amplification. Maternal educational attainment increased over time, in keeping with Australian educational trends.

Age of detection and hearing aid fitting fell significantly over time (Table 3). Thus, children with mild loss detected under Mature UNHS had an estimated diagnosis on average 9 months (95% CI -22.8 to 4.8) and fitting 19 months (95% CI -33.2 to -4.8) earlier than under the Newly Established UNHS system.

Secular trends across the four detection systems (Aim 1): For moderate losses, Figure 2 appears to indicate benefits associated with the shift from Opportunistic through to UNHS systems for expressive language (p for trend .05) and receptive vocabulary (p for trend .06). Receptive language improvement was modest and not statistically significant. The very large stepwise reduction in age of aiding for mild losses across the four periods (Table 3)

was not paralleled by similarly clear upward trends for mild losses, whose expressive language and receptive vocabulary remained around two thirds to one standard deviation below normative means. In this context, an apparent late upswing in receptive language with mature UNHS only (Figure 2) is difficult to interpret. Health-related quality of life and behaviour appeared largely unaffected by system of detection for either group.

Age of detection and language outcomes in the pooled sample (Aim 2): Figure 3a shows a single fractional polynomial line of best fit reflecting the trend in each language outcome by age of detection for the pooled mild-moderate sample, while Figure 3b shows these same trends for the four detection systems overlaid. Language and vocabulary outcomes were higher with detection age before 6 and, unexpectedly, after 30 months, and lower with detection between 6-30 months. However, there was little separation of the lines by era of detection.

The regression analyses (Table 4) show that the benefits of earlier detection were almost wholly experienced by children with moderate, not mild, losses. Compared to a diagnosis under 6 months, children with moderate loss diagnosed between 18-30 months had poorer receptive and expressive language (fully adjusted mean difference -16.7 points, 95% CI -30.3 to -3.0; -27.0, 95% CI -40.2 to -13.7 respectively) and receptive vocabulary (-16.4, 95% CI -28.1 to -4.7). Outcomes for mild losses showed little association with age at detection, other than significantly poorer receptive language when detected between 6-18 months.

Comparison of Mature UNHS and Typically Developing groups (Aim 3): Expressive language was significantly poorer in the mild-moderate combined Mature UNHS than the Typically Developing group (fully adjusted mean difference -8.9 points, 95% CI -14.7 to -3.1). Adjusted receptive language, behaviour and health-related quality of life scores were similar to Typically Developing children (Table 5).

DISCUSSION

This population-level study showed that, over a nearly 20 year period of births, expressive language and receptive vocabulary in children diagnosed with moderate congenital hearing loss improved steadily as hearing detection systems shifted from opportunistic to mature UNHS. While our small sample size means we do not rule out benefit to children with mild losses, they did not show the same clear upward trends.

Our results broadly align with other studies indicating that children with mild and moderate hearing loss continue to have lower than expected language skills, even when identified early (Moeller *et al.* 2015). However, unlike other population level studies (Ching *et al.* 2013; Tomblin *et al.* 2015), we do not observe a clear gradient of worsening language ability with increasing severity of loss. This could reflect both heterogeneity in outcomes of our participants and a narrower range of hearing loss in our study, with its focus on relatively milder losses. We contend, however, that these children are highly relevant because in many countries they are now routinely receiving aids in the expectation of developmental benefits ultimately impacting quality of life.

This study complements the large cross-sectional population-based Hearing in Schools Study (HISS), in which a population sample of over 6000 schoolchildren were screened and those with mild loss had similar outcomes to children with normal hearing on broadly the same language measures used here (Wake *et al.* 2006). In HISS, hearing loss was generally slight or very mild (mean better ear PTA of 22.4 dB HL) and no children with mild losses were aided. This contrasts to the 32.0 dB HL mean PTA in our aided sample of mild losses. Our results suggest that adverse outcomes may accrue in the upper range of mild losses, and/or that children whose hearing loss is detected in the early months or years of life are systematically at developmental risk for reasons not solely due to their hearing loss.

Strengths of this study include the integration of both quasi-experimental and longitudinal approaches that provide convergent findings regarding the possible benefits of earlier diagnosis and amplification (Smithers *et al.* 2015). Our representative, population-derived samples should enhance generalizability and reduce the likelihood of bias. No other study to our knowledge has directly and systematically assessed language using standardized tools across multiple different detection systems in populations that were otherwise similar. Importantly, whilst different measures were necessarily used across time to measure outcomes within the same domain, consistent inter-cohort performance patterns were observed. While the age of participants ranged from 5 to 8 years, the use of standardized measures limited the impact that the different developmental stages of the participants may have had on the findings.

Limitations include the relatively small sample size in each cohort, reflecting all children available with the condition of interest in our state-based registers. We nonetheless generated robust findings of improving secular trend for moderate losses, which are plausible and backed up by our larger pooled analyses. We acknowledge that a larger sample size would have firmed conclusions for the mild group but, unfortunately, to our knowledge no other harmonized cohorts spanning such systems with prospective, same-age standardized outcomes exist internationally nor could now be generated. Our historical cohorts did not record detail on participant recruitment at the hearing loss subgroup level, unlike our thoroughly documented youngest cohort (mature UNHS). How this may have affected generalisability is unknown. Our findings may not fully generalise to children with intellectual disability who were not assessed in the oldest cohort (opportunistic detection). Nonetheless, three-group comparisons including these children did not suggest different conclusions, and their exclusion from the four-group comparisons reduced concerns regarding skewness due to basal scores (Wake *et al.* 2016). As with all population studies from pre-UNHS eras, we cannot be certain that all hearing losses were congenital in two of our four groups. The better outcomes of children detected after age 30 months could be explained by any of chance, late-onset hearing loss being misclassified as congenital, and/or better outcomes leading to later diagnosis (i.e. reverse causation). With all children bar one being hearing aid users, results may not generalise to children with unaided losses. As no information on intervention program enrolment or approach was available, we could not quantify any impact on outcomes. Anecdotally, there has been an on-going trend towards

intervention being offered more frequently and inclusively to milder losses. The fact that this was not mirrored in steadily improving performance is concerning.

Unlike children with moderate losses, it is difficult to conclude that children with mild losses are experiencing clear benefit from the profound shift in practice to earlier detection and aiding. This is even more surprising given that both maternal education levels in the general population and amplification technology improved markedly across the two decades separating the oldest from the youngest children. We consider two possible reasons. Children with mild losses may simply not wear adequately-fitted aids for enough time each day (Fitzpatrick *et al.* 2010; Walker *et al.* 2015), when amplification is subtle and may require prolonged delivery to accrue significant benefit. Alternatively, mild hearing loss could represent "overdiagnosis", defined as identification of a real condition for which treatment does not actually benefit an individual's outcomes (Coon *et al.* 2014). This would imply that these children's developmental deficits might not be attributable solely to their hearing acuity. If so, the decision to amplify mild losses early could represent not only overtreatment (i.e. treatment that cannot deliver benefit) but active harm (costs, burden, stigmatization).

Under current detection practices, large numbers of children born with mild bilateral hearing loss will continue to be identified early. There is a need for carefully constructed and controlled trials that compare systematic hearing aid provision to no provision, and that accurately measure usage. Outcomes should include both specific and broad language

outcomes as well as costs and health-related quality of life. Such trials would need to be of adequate size to explore both the hearing thresholds and the minimum 'dose' (percent of time hearing aids must be worn and quality of fitting) beyond which benefits appear to accrue. Otherwise, it will remain impossible to move to an evidence-based and cost-effective system that optimises management for all children with congenital hearing loss based on need and benefit.

In conclusion, for children born with mild hearing loss we observed limited evidence of benefit to language outcomes as a result of earlier diagnosis and amplification. However, we do observe clear benefits in children born with moderate hearing loss. These results suggest that, despite the best intentions made possible by universal newborn hearing screening, our current approach to treating children with mild loss is not producing consistent improvements across time.

Key messages:

- Universal newborn hearing screening (UNHS) often targets moderate or greater hearing losses, but it also frequently detects mild losses.
- Despite the benefits versus costs/harms of earlier detection of mild losses yet to be established, early amplification for these children has rapidly become standard practice.
- For children with moderate losses, language outcomes have improved as detection has moved from opportunistic to risk factor screening to newly established UNHS and mature UNHS.
- Clear benefits of earlier detection and advanced amplification were not obvious in the measured outcomes for children with mild losses.
- An evidence base for effective clinical care of children with mild losses needs to be established, e.g. using randomised trials of early hearing aid provision.

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Construct	Source and measure	Cohort					Additional information
		TD	Mature UNHS	New Est UNHS	Risk Factor	Opport	
Age at diagnosis	Australian Hearing Database			•	•	•	Age at confirmed hearing loss.
	Victorian Infant Hearing Screening Program Database		•				
Receptive and expressive language	DA: Clinical Evaluation of Language Fundamentals – Fourth ed. (CELF-4)(Semel 2006)	•	•				Receptive language (auditory comprehension) and expressive language (expressive communication) scales. All measures are standardized: normative mean 100, SD 15.
	DA: Clinical Evaluation of Language Fundamentals – Third ed. (CELF-3)(Semel 1995)					•	
	DA: Preschool Language Scale-4. Aust. language adaptation (PLS-4)(Zimmerman 2002)			•	•		
Receptive vocabulary	DA: NIH-adaptive Picture Vocabulary Test (NPVT) (Weintraub *et al.* 2013)		•				NPVT: assessment via automated, adaptive iPad delivery with age-corrected standard scores with mean 100, SD 15.
	DA: The Peabody Picture Vocabulary Test-4 (PPVT-4) (Dunn 2007)			•	•		PPVT: 228 items, standardized: mean 100, SD 15.
	DA: The Peabody Picture Vocabulary Test-3 (PPVT-3) (Dunn 1997)	•				•	
Health-related quality of life	PR: Pediatric Quality of Life Inventory 4.0(PedsQL 4.0) (Varni *et al.* 2003)	•	•	•	•		PedsQL 4.0: 23 items. Total score is sum of physical, social, emotional and school functioning sections. Range: 0=worst health to 100=best possible health.
	PR: Child Health Questionnaire (Landgraf 1996)					•	CHQ: 28 items. Physical and psychosocial summary scores used. Higher score indicates better health.
Behaviour and emotion	PR: Strengths and Difficulties Questionnaire: Australian version for 4-10 year olds (Goodman 2001)	•	•	•	•		SDQ: 25 items. Scales: conduct, emotional symptoms, hyperactivity, peer relationships, pro-social behaviour; Total Difficulties score is sum of all difficulties scales excluding pro-social. Range: 0-40.
	PR: Revised Rutter Parent Scale for School-Age Children (Hogg 1997)					•	Revised Rutter: Emotional, conduct and total difficulties scales. Score ≥13 indicates behaviour problem.

Non-verbal cognition (IQ)	DA: Wechsler Nonverbal Scale of Ability (WNV) (Wechsler 2006) DA: Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler 1999) DA: Wechsler Intelligence Scale for Children (WISC)-Third ed. (Wechsler 1991)	•	•	•	•	•	WNV: Two-subtest version (Matrices and Recognition). Standardized: mean 100, SD 15. Range: 30 to 170. WASI: Two-subtest version (Block design and Matrices). WISC: Perceptual organization index. Higher scores indicate greater cognitive ability.

TABLE 1 Key measures

Abbreviations: TD, Typically Developing; New Est UNHS, Newly Established UNHS; Opport, Opportunistic Detection; DA, Direct Assessment; PR, Parent Response

TABLE 2 Characteristics of the sample

	Typically Developing (n=1217)	Mature UNHS (n=21† qaire) (n=20 ax)	Newly established UNHS (n=41‡)	Risk Factor Screening (n=34□)	Opportunistic Detection (n=50¬)
Child					
Age at assessment in months, mean (SD)	88.2 (2.2)	75.4 (12.3)	65.6 (7.1)	63.0 (3.8)	95.0 (5.7)
Male sex, %	50.5	47.6	56.1	55.9	62.0
Severity – 3 freq dB PTA better ear, mean (SD)					
Initial	-	43.6 (11.3)	43.9 (9.4)	41.0 (11.7)	47.9 (10.0)
Current	-	44.0 (13.6)	50.8 (22.9)	48.0 (26.0)	46.1 (12.0)
Non-verbal IQ*, mean (SD)					
Whole sample	104.7 (14.7)	99.1 (19.2)	94.5 (29.8)	94.1 (25.2)	-
Children without intellectual disability	-	102.9 (9.7)	104.9 (15.1)	100.4 (15.8)	104.6 (18.1)
Family					
Language other than English household, %	5.8	14.3	7.3	11.8	6.0
Disadvantage Index, mean (SD)	1036.0 (60.7)	1007.9 (55.2)	1010.3 (77.1)	1036.8 (50.9)	1016.9 (74.6)
Parent with at least undergraduate level education, %	36.6	57.1	26.8	41.2	32.0

Abbreviations: PTA, Pure Tone Average; qaire, questionnaire; ax, assessment

[*]Non-verbal IQ including un-assessable children for whom the basal standard score (30) was imputed.
[†] Percent missing data (mature UNHS sample): non-verbal IQ 9.1%, all other measures complete.
[‡] Percent missing data (newly established UNHS sample): non-verbal IQ 12.2%, LOTE 2.4%, parent education 9.8%, all other measures complete.
 Percent missing data (risk factor): LOTE 2.9%, parent education 5.9%, all other measures complete.
¬ Percent missing data (opportunistic): LOTE 6%, disadvantage index 2%, parent education 6%, all other measures complete.

TABLE 3 Ages of detection and hearing aid fitting by severity across detection systems (intellectual disability excluded)

Outcome in months	Fully adjusted mean *				Fully adjusted mean difference (95% CI)				
	Mature UNHS (n=18)	Newly established UNHS (n=31)	Risk Factor (n=31)	Opportunistic (n=50)	Mature UNHS	Newly established UNHS	Risk Factor	Opportunistic	P (trend)
Mild									
Age detected (n=56)	4.5	13.5	19.2	31.1	Ref	-9.0 (-22.8 to 4.8)	-14.8 (-27.4 to -2.1)	-26.6 (-39.2 to -14.0)	<.001
Age hearing aid fitted (n=52)	8.3	27.3	21.6	33.3	Ref	-19.0 (-33.2 to -4.8)	-13.3 (-26.3 to -0.3)	-25.1 (-38.1 to -12.0)	.002
Moderate									
Age detected (n=74)	5.5	6.6	21.4	20.8	Ref	-1.1 (-12.8 to 10.5)	-15.9 (-27.4 to -4.5)	-15.3 (-25.4 to -5.1)	<.001
Age hearing aid fitted (n=66)	12.4	8.8	22.5	22.4	Ref	3.6 (-9.7 to 16.9)	-10.1 (-23.4 to 3.1)	-10.0 (-21.6 to 1.6)	.008
Mild Moderate combined									
Age detected (n=130)	5.4	9.9	18.6	25.0	Ref	-4.6 (-13.0 to 3.8)	-13.3 (-21.3 to -5.2)	-19.7 (-27.2 to -12.1)	<.001
Age hearing aid fitted (n=118)	11.2	17.2	20.5	26.8	Ref	-6.0 (-15.3 to 3.4)	-9.2 (-18.4 to -0.1)	-15.6 (-24.1 to -7.0)	<.001

*Adjusted for parent education, English as a second language, disadvantage index, sex, non-verbal IQ and current hearing loss

Percent missing data: Age hearing aid fitted 9.2%, Age detected complete.

TABLE 4 Outcomes by age of detection (in months) for children without intellectual disability

Diagnosis age	Fully adjusted mean by age of detection, with comparisons for later-detected groups against the reference of 0-6 months*											
	0-6 mo	6-18 mo	Mean Diff (95% CI)	P	18-30 mo	Mean Diff (95% CI)	P	30-42 mo	Mean Diff (95% CI)	P	42-54 mo	Mean Diff (95% CI)	P
Mild													
Receptive language	95.7	80.8	-14.9 (-27.1 to -2.7)	.02	84.2	-11.5 (-27.8 to 4.7)	.16	93.5	-2.3 (-15.9 to 11.4)	.74	98.1	2.4 (-11.9 to 16.7)	.74
Expressive language	90.3	84.3	-6.0 (-18.9 to 7.0)	.36	79.1	-11.2 (-28.5 to 6.0)	.19	90.0	-0.5 (-14.9 to 14.0)	.95	91.6	1.3 (-13.9 to 16.4)	.87
Receptive vocabulary	85.3	90.6	5.3 (-6.7 to 17.3)	.38	87.0	1.7 (-14.3 to 17.6)	.84	88.1	2.7 (-10.7 to 16.2)	.68	90.3	5.0 (-9.2 to 19.1)	.48
Moderate													
Receptive language	94.3	85.4	-8.9 (-20.9 to 3.0)	.14	77.6	-16.7 (-30.3 to -3.0)	.02	89.9	-4.4 (-21.1 to 12.3)	.60	88.9	-5.4 (-22.7 to 11.8)	.53
Expressive language	94.9	80.6	-14.3 (-25.9 to -2.7)	.02	67.9	-27.0 (-40.2 to-13.7)	<.001	84.2	-10.7 (-27.0 to 5.5)	.19	82.1	-12.8 (-29.5 to 3.9)	.13
Receptive vocabulary	94.4	85.9	-8.5 (-18.8 to 1.9)	.11	78.0	-16.4 (-28.1 to -4.7)	.007	80.1	-14.2 (-28.6 to 0.1)	.05	90.8	-3.5 (-18.4 to 11.3)	.63
Mild Moderate combined													
Receptive language	93.8	85.8	-8.0 (-16.1 to 0.0)	.05	80.3	-13.5 (-23.4 to -3.7)	.008	91.3	-2.5 (-12.7 to 7.7)	.63	92.2	-1.6 (-12.4 to 9.2)	.77
Expressive language	92.5	84.5	-8.0 (-16.4 to 0.4)	.06	71.9	-20.5 (-30.8 to-10.3)	<.001	86.7	-5.8 (-16.4 to 4.8)	.28	84.7	-7.8 (-19.1 to 3.5)	.18
Receptive vocabulary	90.1	89.2	-0.9 (-8.4 to 6.5)	.80	81.3	-8.9 (-18.0 to 0.3)	.06	84.2	-5.9 (-15.4 to 3.6)	.22	89.1	-1.1 (-11.1 to 9.0)	.83

*Adjusted for parent education, English as a second language, disadvantage index, sex, non-verbal IQ and current hearing loss

TABLE 5 Outcomes by severity for Mature UNHS versus Typically Developing group

Outcome	Normative mean (SD)	Demographically adjusted mean *				Fully adjusted mean †			
		Typically developing	Mature UNHS	Mean Diff (95% CI)	P	Typically developing	Mature UNHS	Mean Diff (95% CI)	P
Mild									
Receptive language	100 (15)	94.3	95.9	1.6 (-7.8 to 11.0)	.74	94.3	101.2	6.9 (-1.9 to 15.7)	.12
Expressive language	100 (15)	98.9	86.1	-12.8 (-21.9 to -3.7)	.006	99.0	91.5	-7.4 (-16.3 to 1.4)	.10
Behaviour problems	6.9 (5.1)	7.0	8.8	1.8 (-1.6 to 5.1)	.30	7.0	9.5	2.6 (-1.0 to 6.1)	.16
Health-related quality of life	81.9 (12.6)	83.4	80.0	-3.5 (-10.6 to 3.6)	.34	83.6	77.7	-5.9 (-13.2 to 1.4)	.11
Moderate									
Receptive language	100 (15)	94.3	95.1	0.8 (-7.7 to 9.3)	.85	94.3	94.9	0.6 (-6.9 to 8.0)	.88
Expressive language	100 (15)	98.9	89.1	-9.7 (-18.0 to -1.5)	.02	99.0	89.0	-10.0 (-17.5 to -2.4)	.01
Behaviour problems	6.9 (5.1)	7.0	8.9	1.8 (-1.1 to 4.8)	.22	7.0	8.4	1.4 (-1.8 to 4.6)	.39
Health-related quality of life	81.9 (12.6)	83.4	86.0	2.5 (-3.6 to 8.7)	.42	83.6	85.5	1.9 (-4.6 to 8.5)	.56
Mild Moderate combined									
Receptive language	100 (15)	94.3	95.5	1.2 (-5.1 to 7.6)	.70	94.3	97.5	3.2 (-2.6 to 8.9)	.28
Expressive language	100 (15)	98.9	87.9	-11.0 (-17.2 to -4.8)	<.001	99.0	90.1	-8.9 (-14.7 to -3.1)	.003

Outcome	Normative mean (SD)	Demographically adjusted mean *				Fully adjusted mean †			
		Typically developing	Mature UNHS	Mean Diff (95% CI)	P	Typically developing	Mature UNHS	Mean Diff (95% CI)	P
Behaviour problems	6.9 (5.1)	7.0	8.8	1.8 (-0.4 to 4.0)	.11	7.0	8.9	1.9 (-0.5 to 4.3)	.11
Health-related quality of life	81.9 (12.6)	83.4	83.3	-0.1 (-4.8 to 4.6)	.97	83.6	82.0	-1.5 (-6.5 to 3.4)	.54

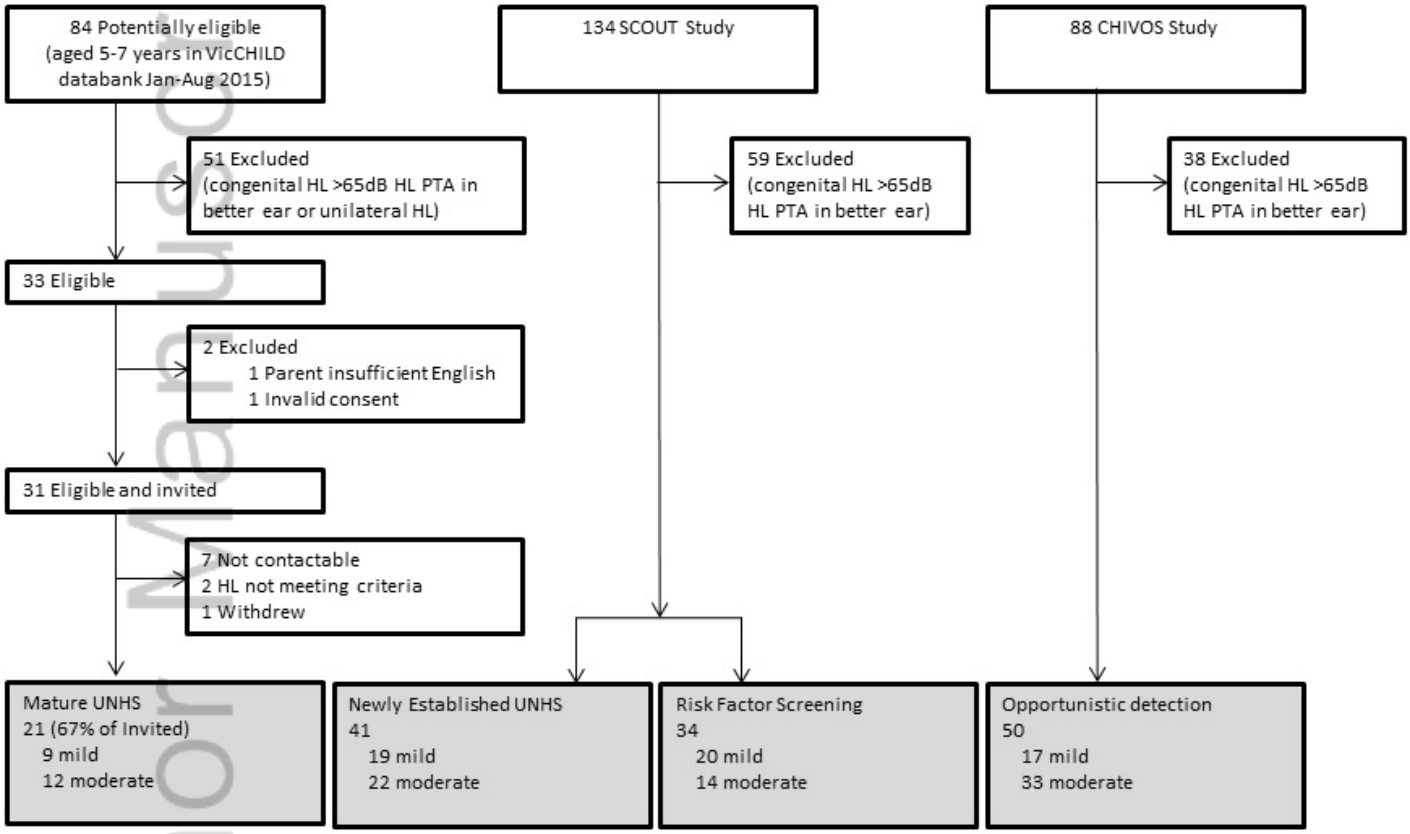
*Adjusted for parent education, English as a second language, disadvantage index, sex †With additional adjustment for non-verbal IQ

Figure 1. Study recruitment from hearing loss cohorts.

Figure 2. Relationships of mean receptive and expressive language and receptive vocabulary outcomes with evolving systems of hearing loss detection.

Figure 2 footnote. Horizontal solid and dashed lines represent normative scores and standard deviations. Whiskers represent 95% confidence intervals. Statistical test of significance, p for trend. Participants with intellectual disability excluded. Abbreviations: Opport., opportunistic; NE UNHS, newly established UNHS; M UNHS, mature UNHS.

Figure 3. Language outcomes by age at diagnosis **(a)** pooled across all systems of detection and **(b)** within detection systems (intellectual disability excluded).



Abbreviations: HL PTA, Pure Tone Average Hearing Loss

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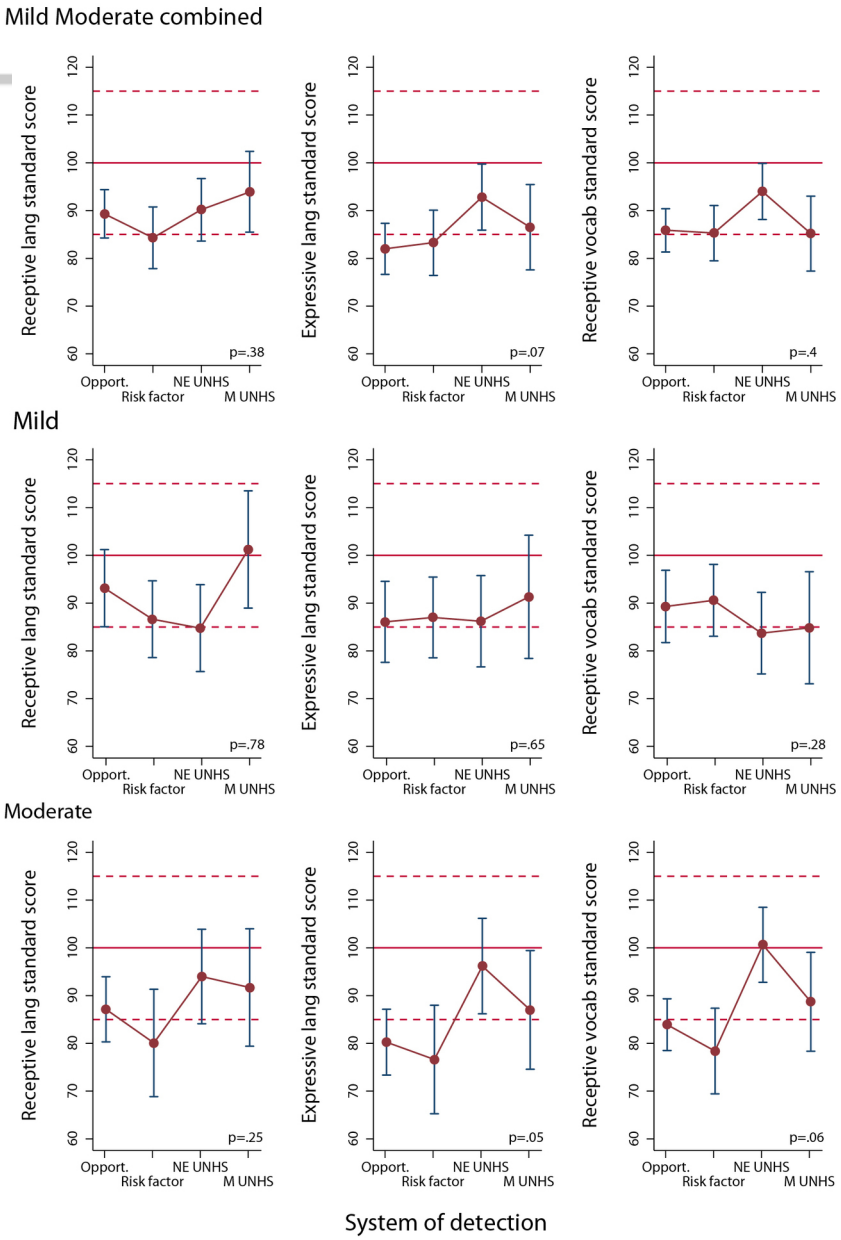

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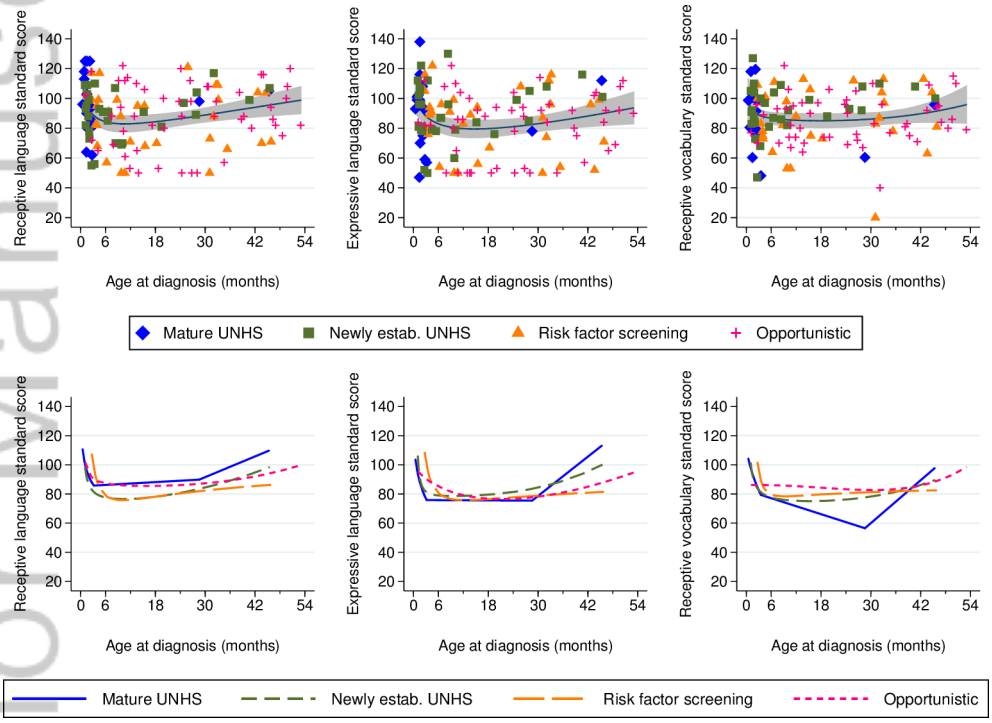

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