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Major findings of the LOCHI study on children at 3 years of age and implications for audiological management

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The Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study addresses the evidence gap regarding the efficacy of universal newborn hearing screening (UNHS) for improving outcomes through a prospective comparison of outcomes of children who were identified either early or late due to differential access to UNHS, but who had uniform access to the same consistent post-diagnostic audiological intervention from a single government-funded national service provider, Australian Hearing. The primary aim was to determine the factors, including age at amplification, that affect outcomes of children with hearing loss.

A major strength of this study was the characteristics of the population-based cohort of 451 children studied. The group was homogenous in terms of age at testing (all were 3 years of age), audiological intervention received (consistently administered according to national protocols, at no cost to families), and onset of deafness (prelingually deaf). The age at hearing aid fitting of the cohort spanned a range that is more in keeping with current practice in countries where UNHS is implemented, unlike those reported in previous studies. Of the children who were referred from newborn hearing screening, the median age of fitting was 3.3 months (interquartile range: 2.2 to 6.5). The cohort was linguistically and culturally diverse. Although the majority of children used spoken English at home, about 28 different languages were also spoken at home. One quarter of the children used signed support to communication at home and/or in their early education setting. The group was educationally diverse, representing children who received early education using communication approaches that ranged from exclusively oral emphasizing spoken language skills to combinations of oral and sign methods to those that emphasised signed communication skills. In drawing from an entire population, rather than from samples self-selecting to obtain services from particular providers, the results reported are not restricted to any particular centre or type of rehabilitation program.

The LOCHI study provided comprehensive data for examining the relationships between different outcomes and predictors, and incorporated randomised controlled trials of hearing

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aid prescription and nonlinear frequency compression. The articles in this supplement reported the findings from these trials, examined the parents' perspectives on early intervention, and the effect of auditory neuropathy spectrum disorder on child outcomes.

This paper presents a summary of the major findings of the cohort evaluated at three years of age. The findings have implications for therapy and expectations for children's development.

Research strategy

The LOCHI cohort comprised 451 children with hearing loss who first received audiological intervention provided by Australian Hearing before they turned three years of age in New South Wales, Victoria and South-eastern Queensland in Australia. As the children had hearing loss ranging from mild to profound degrees, users of hearing aids and cochlear implants are included in the same study. Children from non-English speaking background were included (Crowe et al., 2012; Ching et al., 2013), as were children with additional disabilities (Cupples et al., 2013) and children with auditory neuropathy (Ching et al., 2013).

The test battery was carefully selected to assess the skills that rely on auditory input to develop. These included auditory functional ability, speech production, receptive and expressive language, and psychosocial development at 3 years of age. The measures included tests directly administered to children, and reports solicited from parents or primary caregivers. At later measurement points, nonverbal cognitive ability, literacy, educational attainment, mental health, and quality of life will also be evaluated.

Information about a range of factors associated with the child, family and intervention that have been identified in the literature to potentially influence developmental outcomes were gathered prospectively.

Analysis

In achieving the primary goal of investigating the influence of age of intervention, together with a range of predictor variables, on outcomes, multiple regression analysis was used. The test scores from multiple test measures were combined into a global language score by using factor analysis. The analysis revealed high factor loadings for abilities as diverse as vocabulary, language reception, language expression, and speech production; and gave confidence that the combination of test scores into a global language score was a reasonable representation of the individual assessment tools. This is consistent with the assumption that an overall developmental index underlies the performance in those tests.

Multiple regression analysis was performed with the global language score as a dependent variable, and a range of demographic characteristics as predictor variables. Using the global language score as the dependent variable in the regression has two main advantages. Firstly, the effects of measurement errors and other random variations in individual test scores were reduced in using the underlying global outcomes factor score that was made up of multiple measured scores. Secondly, the global outcomes score allowed a larger sample size than if an individual test scores were used as a dependent variable, because each subject needed a

minimum of two individual scores to have a global score. The use of multiple imputations corrected (in theory) for the uncertainty in the missing scores for some tests. The current approach maximised statistical power in the analysis and generated more reliable estimates than if an individual test score were used in the regression model.

Major findings

Outcomes of children at 3 years of age

The test scores from nine measures were aggregated into a global language score, which was scaled so that a normal population should have a mean of 100 and a standard deviation of 15. The mean global language score for the entire cohort was 74.6 (SD: 17.1). By including children with additional disabilities in the cohort, we found that about 6% of the performance score could be attributed to the presence of other disabilities. After adjusting the scores for the effect of additional disability, the mean global language score was 77.8. This is below 1 SD of the normative mean, revealing the gap between children with hearing loss and those with normal hearing at 3 years of age. Detailed analysis of the outcomes of children with additional disability is reported in a separate study, showing a significant effect of maternal education and severity of hearing loss (Cupples et al., 2013).

Factors influencing global language outcomes

Factors influencing global language outcomes included severity of hearing loss, gender, presence of additional disabilities, gender, and age at cochlear implantation.

Higher performance was obtained for girls than for boys. Gender effects have been documented for language measures of normal-hearing children (Reilly et al., 2010). Gender was also found to be a significant factor influencing young deaf children learning oral language in auditory-verbal programs (Easterbrooks & O'Rourke, 2001); and school-aged children with cochlear implants (Geers et al., 2003). The finding suggests that this characteristic of children needs to be controlled in future research on speech and language outcomes of children with hearing loss.

Maternal education was a significant predictor of language outcomes at 3 years – a finding that is consistent with reports on language development of normal-hearing children at 2 and 4 years of age (Reilly et al., 2010). Global language scores were higher for children of mothers who completed university education compared to those of mothers who completed ≤12 years of schooling (effect size = 6.3 points, +/− 4.3). The effect of maternal education may be related to the quality and quantity of communicative input received by children in their home environment (Quittner et al., 2013).

For children using cochlear implants by 3 years of age, higher performance was associated with earlier implantation. Delaying implantation from 12 to 24 months of age was associated with a reduction of global outcomes score by about 0.5 SD. Previous studies that compared children who received an implant before or after 5 years of age documented auditory and speech advantage for children implanted before 5 years of age (Moog & Geers, 2003; Fink et al., 2007; Clark et al., 2012). They also raised uncertainties about why outcomes were not affected by when cochlear implantation was provided within the first 5 years of age. No

previous studies investigated the effect of age of implantation in a population who received a cochlear implant before 3 years of age, spanning a range of 5 to 30 months of age that is more consistent with current practice. In the present cohort of 134 children, the effect of age of activation of cochlear implants was significant after controlling for the effects of a range of predictor variables. Furthermore, this study found that global language scores for children with bilateral cochlear implants were higher than those with bimodal fitting (6.6, +/− 7.2) or those with unilateral cochlear implants (8.9, +/− 10.6); although the differences did not reach significance level (Ching et al., in press). The findings support the provision of auditory stimulation in both ears, either through electrical stimulation or acoustic stimulation, or a combination of both from an early age.

Effect of hearing aid prescription

By randomly assigning newly identified children to one of the two prescriptions for fitting, this trial produced high-level evidence showing that the choice of prescription did not have a significant effect on global language outcomes scores at 3 years. Although the estimated effect size is positive, it is only 1.0 point with a confidence interval of +/− 3.2 points. This suggests that the true mean effect lies between −2.1 points and 4.2 points. There were no significant effects of choice of prescription on children's language, speech production or functional performance in real life (Ching et al., in press).

This finding is consistent with theoretical modelling that revealed no significant difference in estimated speech intelligibility between prescriptions (Ching et al., in press). For a certain audiogram, the gain differences between prescriptions resulted in greater loudness for the DSL than for the NAL prescription. Contrary to assumptions that threshold shifts are unlikely when hearing aids with wide-dynamic range compression are used, our calculations indicated that use of hearing aids in listening environments of high sound pressure levels would lead to predicted threshold shifts for hearing loss exceeding about 70 dB HL when DSL is used, and about 90 dB HL when NAL is used (Ching et al., in press).

Effect of nonlinear frequency compression

The trial revealed that an effect size close to zero (0.8 global language score points) was associated with the use of nonlinear frequency compression (NLFC) relative to conventional amplification for children at 3 years of age (Ching et al., in press). With a confidence interval of +/− 7.5 points, the true mean effect of using NLFC lies within the range of −6.7 points to +8.3 points. A larger sample size may reveal clinically significant differences, in either direction, between NLFC and conventional processing. Current evidence is insufficient to reject the null hypothesis that there is no difference in language outcomes between the device types, for children with audiometric characteristics like those in the present study, which fit within manufacturer-specified fitting range for NLFC in hearing aids. Speech production data revealed an increase in erroneous substitution of affricates by fricatives for children who used NLFC compared to children who used conventional amplification. Given that young children rely on the auditory input to develop speech and language, their speech perception is more susceptible to spectral distortions, such as NLFC, than are older children or adults; these findings have important implications.

Effect of the presence of auditory neuropathy spectrum disorder

The presence of auditory neuropathy was not a significant factor affecting outcomes, after allowing for the effects of hearing sensitivity and a range of demographic characteristics in the regression model (Ching et al., 2013). This finding applied for children with hearing aids and children with cochlear implants. None of the previous studies adjusted for the range of variables we included in the regression model to estimate the effect of the presence of auditory neuropathy. Although the estimated effect size is positive, it is very close to zero. The mean effect size is only 1.0 point, and the confidence interval is +/− 5.7 points. All we can conclude from the current data is that the true mean effect of auditory neuropathy lies within the range − 4.7 points to + 6.8 points.

Parents' perspectives

A small group of parents expressed strong support for early intervention, and high expectations for their child's development (Gilliver et al., in press). Nonetheless, they also reported anxiety and concerns arising from observations of the difficulties experienced by their child in real life despite consistent device usage. They also expressed the need for information tailored to individual needs.

Clinical implications

The factors contributing significantly to outcomes of children lend support to the impetus for implementing family-centred practice principles in early intervention (Joint Committee on Infant Hearing, 2013; Moeller et al., 2013). Parents articulated the desire for clinicians and professionals to provide them with information and ongoing support that are tailored to their individual needs.

The findings from the randomized trial of hearing aid prescription revealed that choice of prescription does not produce a significant impact on language development or predicted speech intelligibility. However, its influence on loudness and hearing aid safety must form part of an evidence-based guideline for clinical management. Frequent occurrence of loudness discomfort will influence usage of device, and frequent occurrence of temporary threshold shift reduces speech reception ability of the individual child. Regular occurrence of threshold shift that exceeds the safety limit is likely to result in permanent threshold shift. With each increase in hearing threshold is an increase in gain and likely increase in noise exposure over the lifetime of hearing-aid use by a child diagnosed with hearing loss early in life. The findings caution clinicians to be vigilant about potential threshold shifts arising from hearing aid use, and to counsel parents about noise-induced hearing loss for children.

The findings from the randomized trial of nonlinear frequency compression do not lend support to the current practice of activating nonlinear frequency compression in hearing aids for up to 80% of pediatric fittings (Jones & Launer, 2010), and highlight the potential risk for atypical development of speech production (and perception) in young children.

How generalizable are the findings?

UNHS would not be expected to be beneficial without timely and quality auditory intervention and educational support. Given that initiation of intervention begins with hearing aid fitting for almost all children with permanent childhood hearing impairment, any evaluation of the efficacy of UNHS for improving long-term outcomes must necessarily ensure that results are not confounded by variations in post-diagnostic auditory intervention. By taking advantage of the unique situation in Australia where timely and adequate hearing services are provided to all children with hearing loss by the same national service provider, the LOCHI study sampled from an entire population and controlled for consistency of hearing services for all children in investigating the efficacy of UNHS. The findings would be generalizable to other populations.

Limitations

The articles in this supplement reported outcomes of children at 3 years of age. By necessity, the LOCHI study included only children whose hearing loss was detected before 3 years of age. Given the young age, non-verbal cognitive ability could not be reliably measured, and was thus not included as a predictor variable. Nonetheless, the longitudinal nature of the LOCHI study means that the cohort will be assessed at 5 years of age on an increased range of outcomes domains and on cognitive ability. The data at 3 years of age form the basis for future investigations of rate of development of children over the first 5 years of life.

Conclusion

The data from this study are yielding important information that support the effectiveness of early detection for improving child language at 3 years, but also reveal deficits that require targeting with improved intervention practice. While the data to date are already valuable, continuing to monitor children’s outcomes at an older age will allow assessments of critical literacy skills that cannot be measured at a young age. Such measurements are necessary to investigate whether UNHS is effective in achieving its ultimate goal of optimising language, social, and literacy development for children with permanent childhood hearing loss.

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