Learning from the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study: summary of 5-year findings and implications

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Key words: pediatric, hearing aid, cochlear implant, outcomes, early intervention, LOCHI study

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Abbreviations:

AH: Australian Hearing

C-levels: Comfortable levels

CI: Cochlear implant

DSL: Desired Sensation Level procedure

HA: Hearing aid

LOCHI: Longitudinal Outcomes of Children with Hearing Impairment

NAL: National Acoustic Laboratories

PCHL: Permanent childhood hearing loss

PEACH: Parents' Evaluation of Aural/oral performance of Children

rms: Root-mean-square

T-levels: Threshold-levels

UNHS: Universal newborn hearing screening

Abstract

Objective: This article summarises findings of the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study, and discusses implications of the findings for research and clinical practice.

Design: A population-based study on outcomes of children with hearing loss. Evaluations were conducted at five years of age.

Study sample: Participants were 470 children born with hearing loss between 2002 and 2007 in New South Wales, Victoria and Queensland in Australia, and who first received amplification or cochlear implantation by three years of age.

Results: The earlier hearing aids or cochlear implants were fitted, the better the speech, language and functional performance outcomes. Better speech perception was also associated with better language and higher cognitive abilities. Better psychosocial development was associated with better language and functional performance. Higher maternal education level was also associated with better outcomes. Qualitative analyses of parental perspectives revealed the multiple facets of their involvement in intervention.

Conclusions: The LOCHI study has shown that early fitting of hearing devices is key to achieving better speech, language and functional performance outcomes for children with hearing loss. The findings are discussed in relation to changes in clinical practice and directions for future research.

Introduction

The Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study is a prospective, quasi-experimental investigation of the effects of age at intervention and other factors on outcomes of children with permanent childhood hearing loss (PCHL) (Ching et al., 2013d). The study increases understanding about the impact of PCHL on developmental outcomes in a population-based sample, and generates evidence that guides improved management of PCHL to optimise child outcomes.

The research team enrolled about 470 children born with hearing loss between 2002 and 2007 in New South Wales, Victoria, and Queensland, and who first received hearing aids or cochlear implants younger than three years of age. Some children had access to universal newborn hearing screening (UNHS) whereas others not, depending on whether UNHS was operating in their region of birth. The study controlled for variations in post-diagnostic audiological management, with all children receiving the same consistent post-diagnostic services from Australian Hearing (AH), the government-funded organization that provides hearing services to children with hearing loss in Australia.

Evaluations of the outcomes of the cohort in the study were carried out at several time points. Findings on the outcomes at three years of age have been reported previously (Ching & Dillon, 2013; Ching et al., 2013c). In the collection of articles in this Supplement, detailed reports on the perspectives of parents about diagnosis and intervention have been provided, comprehensive descriptions about the characteristics of hearing aids and cochlear implants worn by children have been included, and investigations of factors influencing a range of outcomes, including language, functional performance, speech perception, and psychosocial

skills, have been reported in detail. This paper summarises results across the various outcomes domains and considers the practical implications of these findings for management of children with PCHL.

Summary of findings

Parents' perspectives about diagnosis and intervention

About 85% of parents/caregivers who completed a questionnaire survey reported that they were satisfied with the information and emotional support provided to them after diagnosis of their children's hearing loss (Scarinci et al., this issue). However, a few families indicated that they experienced a breakdown in information transfer with health professionals, and reflected on the post-diagnostic period as a difficult and emotional time. They described the process of accessing intervention services for their child as navigating a maze.

Parental involvement in early intervention was further explored through semi-structured interviews (Erbasi et al., this issue). Parents perceived themselves as central to the intervention of their children and held themselves responsible for their children's outcomes. They served multiple roles including those of a case manager in organising multiple appointments and attending them with the child while making arrangements for the family, a care-provider in making sure that the child is well prepared before attending appointments, a teacher, and an advocate for the child's needs. Parents noted that they always have the best interest of the child's language development in mind in their daily lives, and amidst the multitasking, they are parents. They considered that the multi-faceted nature of their involvement in their children's intervention has not always been recognised by professionals. These findings suggest that it is important for service providers to acknowledge the many roles that

parents play and provide them with support to fulfil those roles. It is also important for service providers to not reduce the parent-child relationship into a pedagogical relationship (Suissa, 2006). Given that parents held themselves accountable for their child's outcomes and experienced a sense of failure when the child did not appear to progress, it would be useful to offer counselling services to reduce the risk of parents experiencing guilt or other negative emotions. The findings are consistent with, and lend support to, the recommendations outlined in the international consensus statement on family centred early intervention (Moeller et al., 2013).

Audiological intervention: HA fitting

Children in the LOCHI study appeared to have benefited from the presence of a national government-funded organisation that provides consistent audiological management and technology to all children with PCHL. The study showed that:

- Device fitting occurred shortly after diagnosis, regardless of whether the hearing loss was diagnosed via UNHS or standard care, or the geographical state of residence. Comparison of the medians of the time gap between diagnosis and first fitting for participants in the LOCHI study showed no significant difference between screened and non-screened groups. The median ages at fitting of hearing aids (HAs) were 3.5 months (interquartile range: 2.3-7.3) and 16.4 months (interquartile range: 7.2-25.8) for the respective groups.
- Measurements of HAs used by children in the LOCHI study revealed that prescriptive targets were met within 3 dB root-mean-square (rms) error across the range from 0.5 to 4 kHz; both when measured at three years of age (Ching et al., 2013b) and at five years of age (Ching et al., this issue). A close proximity to targets was achieved,

regardless of whether the National Acoustic Laboratories (NAL) or the Desired Sensation Level procedure (DSL) prescription was used for HA fitting.

The randomised trial of NAL and DSL prescriptions revealed no significant differences between prescription groups in language, speech production and speech perception scores at five years of age (Ching et al., this issue). When HAs matched targets of the respective prescriptive procedures that are supported by good empirical evidence, they enable the same speech intelligibility and language development. The findings lend support to the American Academy of Audiology guidelines for best practice in pediatric amplification (American Academy of Audiology Task Force on Pediatric Amplification, 2013).

Audiological intervention: CI characteristics

All children who wear cochlear implants (CIs) use the Cochlear Nucleus devices. They received programming services from different CI service centres across the three states, while continuing to receive hearing-related services from AH including but not limited to remote microphone systems, upgrades of CI processors, and accessories. We found that:

- Clinical practice is consistent across sites, with most CIs being programmed according to default values recommended by the manufacturer. The proportion of non-default settings in pulse width, stimulation rate, number of active electrodes and number of maxima was higher in children with auditory nerve deficiency or cochlear lesions, compared to children without those conditions (Incerti et al., this issue).
- Children with cochlear structural and/or neural lesions required significantly higher current levels for threshold and comfortable levels (T- and C-levels), as compared to children without those conditions. Hence, approaches for programming CIs for

children with cochlear structural and/or neural lesions need to be less reliant on interpolation of levels or global adjustment techniques than those for children without those conditions.

- Averaged across all children, C-levels at six months after activation of their CIs were significantly lower than those at three and five years.
- Averaged across all children, there were no significant differences between three and five years of age in T-levels, C-levels, and dynamic range in CIs worn by children.
- Comparing the CI settings in children who first received their CIs by 12 months of age (early-implanted) to those who received their CIs between 12 months and three years (later-implanted), we found that T-levels were significantly higher and dynamic ranges were significantly narrower for the early-implanted group. These findings, discussed in detail in Incerti et al (this issue), call for new programming tools to improve clinical practice so that children who receive CIs earlier gain access to a wider range of sounds earlier to capture the benefits of early implantation.

Factors influencing outcomes: early fitting of HA or CI is key

Children's language, functional performance, speech perception, and psychosocial skills were measured at five years of age. The findings show that the earlier the intervention commenced, the better were the outcomes. This result applies over the entire range of intervention ages measured, which is 0.9 to 35.8 months for children wearing HAs and 5.3 to 35.3 months for children wearing CIs. The benefit of early fitting is greater for those with poorer hearing (Ching et al., 2017a).

- Earlier device fitting (HA or CI) was associated with higher global language scores (summarising language ability, speech production and speech perception evaluated

using a range of measures). For those with HAs, the impact of later fitting increased with the degree of hearing loss (Ching et al., 2017a).

- Earlier device fitting (HA or CI) was associated with better receptive and expressive language.
- Higher nonverbal cognitive ability was associated with better receptive and expressive language, speech production, and functional performance in everyday life (Cupples et al., this issue-b).
- For children wearing HAs, less severe hearing loss and higher levels of maternal education were also significantly associated with better language outcomes (Cupples et al., this issue-b).
- The absence of additional disabilities was significantly associated with better language outcomes for children using CIs, but not for those using HAs (Cupples et al., this issue-a).
- A randomised controlled trial of the NAL and DSL prescriptions showed that there were no significant between-group differences in language or speech perception outcomes (Ching et al., this issue).
- For children with additional disabilities, better language outcomes were associated with earlier fitting of HAs, lesser hearing loss, higher cognitive ability, use of speech for communication, and higher level of maternal education (Cupples et al., this issue-a).
- On average, children with PCHL required a better signal-to-noise ratio than their peers with normal hearing to achieve the same level of performance for speech perception in noise (Ching et al., 2017b).

 On average, children with PCHL demonstrated spatial release from masking for speech perception in noise that is similar in magnitude to that of their peers with normal hearing (Ching et al., 2017b).

- Children who had better language abilities also had better speech perception in noise (Ching et al., 2017b).
- Earlier activation of CIs was associated with better speech perception in noise (Ching et al., 2017b).
- Psychosocial skills as rated by parents showed that better performance was associated with better language ability and functional performance skills as measured by the Parents' Evaluation of Aural/oral functional performance of Children (PEACH; Ching & Hill, 2007) scale at the same age (Wong et al., this issue).

Discussion

The findings of the LOCHI study attest to the importance of early fitting of HAs or CIs soon after diagnosis. Table 1 gives a summary of these findings together with clinical implications.

Table 1 about here

It is noteworthy that consistent usage of hearing devices has been established at an early age in the present cohort. Based on parents' ratings, 62% of the cohort used their devices for more than 75% of their waking hours by three years of age (Marnane & Ching, 2015). This proportion increased to 85% by five years of age.

Incorporating knowledge into clinical practice

The LOCHI study showed that language delay in children with PCHL is abatable, or in some cases, completely preventable. The earlier a child receives HAs or CIs, the better are the language and speech perception outcomes at five years of age. With the widespread implementation of UNHS, it is now possible for PCHL to be identified soon after birth. In order for timely amplification to occur soon after diagnosis, a seamless clinical pathway from screening to diagnosis to early fitting of devices is essential. Current practice in Australia requires that AH provides a post-diagnostic appointment at a hearing centre to a child diagnosed with PCHL at primary healthcare centres within 10 working days. The median HA fitting age of 3.5 months for those children in the LOCHI cohort who had access to UNHS is consistent with national fitting statistics (Australian Hearing, 2017).

The LOCHI study has shown that HAs fitted to children provide consistent audibility to support speech and language development (Ching et al., this issue). These children received post-diagnostic services from AH according to national protocols (King, 2010; Punch et al., 2016) that incorporated evidence-based guidelines for pediatric amplification as published by the American Academy of Audiology (American Academy of Audiology Task Force on Pediatric Amplification, 2013). In a similar vein, Bagatto et al (2016) have reported on clinical feasibility of adopting the guidelines using specific protocols in the Ontario Infant Hearing Program, and showed that children wearing HAs achieved good outcomes when the protocols were executed.

Further, children who need CIs must receive them early to achieve the best language and speech perception outcomes. This, together with findings about the predictability of language scores from early PEACH scores (Ching et al., 2013a), have contributed to national protocols

for pediatric referral for cochlear implantation and monitoring progress after amplification in Australia (Ching et al., 2008; King, 2010). The protocols emphasise the need to evaluate the effectiveness of HAs for infants by using objective measurements of cortical auditory potentials evoked by speech at conversational levels and subjective parent reports using the PEACH scale (Ching & Hill, 2007) as part of routine clinical practice (Punch et al., 2016).

The LOCHI study provides some evidence to support the use of the PEACH scale as a clinical tool for evaluating the effectiveness of amplification in infants. Data from evaluation of the cohort at 3 years of age showed a significant positive relationship between PEACH scores based on parent ratings and standardised language scores measured by administering the Pre-school Language Scale (Zimmerman et al., 2002) directly to children at the same age (Ching et al., 2010). Furthermore, earlier PEACH performance evaluated at either 6 or 12 months after HA fitting was found to be a significant predictor of language outcomes measured at 3 years of age, after allowing for the effects of a range of demographic characteristics in the LOCHI study (Ching et al., 2013a).

The evidence suggests that the PEACH scale can be used as a means to monitor language development of children with hearing loss. The PEACH takes less than 15 mins to complete, whereas a standardised language test based on either parent reports or direct administration to a child requires much longer to complete. The PEACH specifically asks parents to observe and rate their child's listening and communication skills in both quiet and noisy situations in real life, reflecting pragmatic aspects of spoken language and auditory behaviour. On the other hand, standardised tests are typically administered in ideal listening environments, viz, one-on-one in quiet. The PEACH can be administered by audiologists or other healthcare

professionals, and does not require specific expertise in speech and language assessments. Further, the PEACH scale has the benefit of being suitable for use with young children and those who cannot complete standardised testing. As the scale is available in a range of languages (freely downloadable from www.outcomes.nal.gov.au), it can also be used with families from non-English-speaking backgrounds. When used as part of routine management of infants under three years of age, the PEACH scale can be administered within a couple of months after initial HA fitting, and subsequently at 6-monthly intervals to track progress.

In addition, the PEACH scale can be a useful screening tool for identifying psychosocial deficits in young children. This is supported by findings on children in the LOCHI study at three years (Leigh et al., 2015) and at five years of age. Wong et al (this issue) showed that even after accounting for demographic characteristics and language ability, functional communication as measured by the PEACH scale accounted for significant variance in psychosocial skills evaluated using standardised methods at five years of age.

Language ability was significantly associated with speech perception (Ching et al., 2017b) and psychosocial abilities (Wong et al., this issue). The direction of causation cannot be inferred from our results, but it seems likely that language ability both enables and is enabled by, good speech perception. The same may well be true of psychosocial development if children with closer to normal psychosocial development engage in more interactions with their peers and families than those with poorer psychosocial development (Fellinger et al., 2009; Dammeyer, 2010).

Despite early fitting of hearing devices, children with PCHL require higher signal-to-noise ratios compared to their peers with normal hearing to achieve the same level of performance in speech perception (Ching et al., 2017b). Presumably, this is because of the degraded analytical ability of a damaged cochlea, which amplification cannot correct. This finding supports the provision of hearing technology that not only increases audibility but also improves signal-to-noise ratio to children with PCHL. Current AH protocols recommend the fitting of HAs with directional microphone technology and remote microphone systems to young children.

The report on children with disabilities in addition to PCHL showed that early amplification led to improved language outcomes (Cupples et al., this issue-a). Therefore, their access to early fitting of hearing devices should not be compromised. It is important to acknowledge, however, that children with additional disabilities, comprising about 37% of those with PCHL, will need extra support to optimise their language and other outcomes, support that will undoubtedly vary from child to child. Consistent with this view, previous findings for the LOCHI sample revealed that children with autism, cerebral palsy, and/or developmental delay differed from those with other disabilities (which included visual or speech impairment, syndromes not entailing developmental delay, and medical conditions) in attaining poorer language outcomes at three years of age (Cupples et al., 2014); and showing a relative decline in language growth compared to norms from three to five years of age (Cupples et al., submitted). Findings such as these underscore the importance of establishing effective collaborations among professionals in the management of children with hearing loss who have additional disabilities in order to facilitate early treatment for hearing loss.

Future research

We have learnt that the earlier a child receives a CI, the better the language and speech perception outcomes. We have not yet found any limit to the age range over which this is true: language outcomes at five years of age monotonically increase as the age of implantation decreases, at least down to five months of age, the earliest age of implantation in our data (Ching et al., 2017a). Whereas the referral for CI candidacy maybe straightforward and well supported by evidence for children diagnosed with PCHL of a profound degree, referrals are more variable and evidence is less clear for those with hearing loss in the moderate to severe range. Currently, the decision to implant early in infants and young children is made almost exclusively on the basis of threshold elevation and its consequences for detection of speech sounds with amplification (Lovett et al., 2015). For adults and older children, by contrast, the decision for implantation relies almost exclusively on the speech perception ability of the patient when wearing HAs (Gifford et al., 2010; Leigh et al., 2011), as that has been found to be a more reliable indicator of implantation benefit than just hearing thresholds. Clinical tools that enable clinicians to assess auditory speech discrimination in infants are lacking.

Recent research has shown that measurements of objective auditory evoked potentials could reveal speech discrimination abilities in infants and young children (Cheour-Luhtanen et al., 1995; Cone, 2015; Small et al., 2017). Other research groups have demonstrated the use of behavioural measures of discrimination by using a visual reinforcement paradigm (Uhler et al., 2015). Our current research builds on these approaches, with the ultimate goal of developing clinical tools to identify infants with hearing loss who may have deficits in speech discrimination despite optimal amplification. It would then be possible for families to

consider alternative treatment early, with the potential of enabling the infants to benefit from the earliest possible cochlear implantation.

Last but not least, the LOCHI study found that better language outcomes were associated with less severe hearing loss, higher nonverbal cognitive ability, absence of additional disabilities, use of speech for communication, and higher maternal education. The advantages of parental education for child development have been well documented (Bornstein et al., 2010). There is some evidence to suggest that maternal education is related to language input and language environment (Hoff, 2003; Dwyer, 2017), engagement in interaction (Lam & Kitamura, 2012), emotional well-being (Sarant & Garrard, 2014), perceived social support (Ahlert & Greeff, 2012), and self-efficacy (DesJardin, 2005). Future research into how better to support language development in children with PCHL, especially but not limited to those whose mothers did not complete university education or from lower socio-economic backgrounds, is essential (Lam-Cassettari et al., 2015). To enable children with PCHL to achieve parity of outcomes with their normal-hearing peers, further research is needed to increase understanding about how spoken language can be best learnt in the presence of hearing loss, and what are the most effective ways for parents to promote communication.

Conclusion

The LOCHI study has shown that early fitting of hearing devices is key to achieving better speech, language and functional performance outcomes by five years of age. Better language and functional performance are associated with better speech perception and psychosocial development. The longitudinal nature of the study provides the opportunity to track

development over time, and to investigate factors, including but not limited to age at intervention, that influence outcomes at different ages as well as rate of development.

Acknowledgements and Declaration of Interest

Conflicts of interest

None were declared.

Sources of funding

The LOCHI project is partly supported by the National Institute on Deafness and Other Communication Disorders (Award Number R01DC008080). The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institute on Deafness and Other Communication Disorders or the National Institutes of Health.

The work was also partly supported by the Commonwealth of Australia through the Office of Hearing Services. We acknowledge the financial support of the HEARing CRC, established and supported under the Cooperative Research Centres Program of the Australian Government. We also acknowledge the support provided by the New South Wales Department of Health, Australia; Phonak Ltd; and the Oticon Foundation.

Acknowledgements

We gratefully thank all the children, their families and their teachers for participation in the LOCHI study. We also acknowledge the assistance of the many clinicians who provided audiological management of the children, and the assistance of professionals at early intervention agencies and cochlear implant clinics in the collection of demographic and outcomes data. We thank Vicky Zhang for her assistance in the preparation of the manuscript.

Table 1. Summary of findings and implications.

	Research findings	Implications for Practice
Scarinci et al.	Parents were satisfied with support	Intervention programs can do better
(this issue)	and information received after	by
	diagnosis from their rehabilitation	- attending to individual needs;
	audiologists and teachers.	- providing additional support
	Some families expressed the need for	and information for accessing
	extra help and support.	services
Erbasi et al.	Parents play a central role – work	Professionals need to
(this issue)	behind the scenes so that their child	- acknowledge the multiple
	is prepared. They act as case	roles and show sensitivity to
	managers to manage appointments,	parents' involvement behind
	teachers to encourage language	the scenes,
	development, advocates for their	- respect parents' primary role
	child's needs, and want to be parents.	as parents; and
		- empower parents by
		providing support so that they
		can fulfil the multiple roles.
Cupples et al.	Early fitting of hearing aids is	Streamline clinical pathway from
(this issue-b)	associated with better language and	screening to diagnosis to device
	functional performance.	fitting.
Cupples et al.	Early cochlear implantation is	Evaluate effectiveness of
(this issue-b);	associated with better speech	amplification and monitor outcomes

Ching et al.	perception, language, and functional	so that those who need cochlear
(2017b)	performance.	implants can be referred at an early
		age.
Ching et al.	Evidence-based protocols for hearing	Consistent protocols and clinical
(this issue)	aid fitting provided consistent	support to promote adoption of
	audibility when executed – hearing	evidence-based guidelines for
	aids met prescriptive targets within	achieving good fitting outcomes.
	3dB root-mean-square (rms) error.	
Incerti et al.	For children who use cochlear	Improved fitting tools to enable
(this issue)	implants,	children who received cochlear
	- no significant difference in	implants before 12 months of age to
	threshold and comfortable	achieve optimal settings earlier.
	current levels between 3 and	
	5 years of age.	
	- For children without cochlea-	
	vestibular abnormalities,	
	those implanted before 12	
	months had higher threshold	
	levels than those who were	
	implanted after 12 months;	
	when measured at 6 months	
	after CI activation.	
Wong et al.	PEACH is a concurrent predictor of	Use PEACH as a clinical tool for
(this issue);	language and psychosocial	monitoring children's progress in
	outcomes.	routine management.

Ching et al.	Early PEACH at 6 or 12 months after	
(2013a)	fitting is a predictor of 3-year	
	language outcomes.	
Ching et al.	Despite early fitting, children with	- Fit hearing devices with
(2017b)	hearing loss need better signal-to-	technology to improve SNR
	noise ratios (SNR) than their peers	in everyday listening;
	with normal hearing. However,	- Training to integrate audio
	binaural unmasking was of a similar	with visual cues may support
	magnitude between groups.	children who experience
		difficulties listening in noise.
Cupples et al.	Children with hearing loss who have	Presence of additional disabilities
(this issue-a)	additional disabilities benefit from	should not be a reason for delaying
	early fitting of hearing devices for	amplification for children with
	language development.	hearing loss.
Cupples et	Higher maternal education was	Intervention that encourages in all
al., (this	associated with better language	families the communication
issue-a, this	outcomes.	behaviour that presumably tends to
issue-b)		occur more naturally in families with
		higher maternal education level
		would likely improve language
		outcomes.

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