# Exploring the dynamic changes in factors affecting speech and language outcomes in paediatric cochlear implant users

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## Abstract

Multiple factors affect long-term speech and language outcomes in children growing up using cochlear implants (CIs). Influential factors can change over time due to alterations in clinical practice (e.g. updated clinical guidance on indications, introduction of neonatal screening or introduction of meningitis vaccine) or due to developments in society (e.g. population migration affecting cultural/ethnic distribution or socioeconomic changes). If this complex and dynamically changing landscape of influential factors is well understood appropriate interventions can be introduced for families that are most in need of them to facilitate faster rates of language acquisition.

In this research we monitored English speech and language outcomes for children receiving a CI at a central London implant programme from January 1998 to December 2019 with the intention of understanding the most important factors affecting speech and language development and whether these factors changed across the 20-year period. There were 418 children implanted, of which 173 had 5-year post implant speech and language scores. We monitored the distribution of type of onset of loss (congenital, progressive or acquired), age at implantation, home language, socioeconomic status, implant status, core language scores (Clinical Evaluation of Language Fundamentals (CELF) Edition 4). The findings were interpreted with consideration of clinical and societal moderators than can affect outcomes.

In terms of major changes in the population that occurred across the 20-year period we observed an increase in the proportion of children that were implanted with progressive losses. This was most likely affected by the introduction of the UKs National Institution of Health and Care Excellence (NICE) guidance on cochlear implants in 2009, which saw a reduction in the audiometric indication levels such that more children with progressive losses were eligible. There was also an increase in the number of children receiving simultaneous bilateral implants because of that NICE guidance. One of the most striking changes was the increase in the proportion of families who were not native English speakers (75% native English speakers in early years, dropping to 40% in later years), influenced by a change in the migration patterns in the population. There was a reduction in the Index of multiple deprivation (IMD; measure of socio-economic status) with time.

The findings indicated that the majority of children (65%) had severe language delays or were unable to be tested. This figure is often not represented in studies where the inclusion criteria are restricted.

A multinomial logistic regression for core language indicated that the best predictive model comprised of type of onset of severe-to-profound deafness (congenital, progressive, acquired),

implant status (unilateral, bimodal, sequential bilateral, simultaneous bilateral), home language (native English, non-native English), and parental education level.

# Introduction

Cochlear implants (CIs) are one of the most successful yet expensive medical devices available, with over 800,000 recipients worldwide. They are offered to babies, children and adults who have severe-to-profound sensori-neural hearing loss and do not benefit from conventional hearing aids.

For infants, and children CIs have the potential to provide access to sound to facilitate the development of spoken language, good communication and therefore good education outcomes (Niparko 2010). In fact, the evidence that supports this claim is unquestionable and many researchers have observed large improvements (on average) for the development of speech perception, speech production and spoken language skills (Manrique 2004, Dunn 2013, Dettman et al. 2021). However, there is a large variability in outcome and there are a subset of recipients who do not achieve outcomes in the typical age-appropriate range and perform below expectations (Pisoni et al 2017). The size of this poorer achieving population is not well understood.

Multiple factors affect the long-term speech and language outcomes in children growing up using CIs. Influential factors can change over time due to alterations in clinical practice or due to changes in society. If this complex and dynamically changing landscape of influential factors is well understood appropriate interventions can be introduced for families that are most in need of them to facilitate faster rates of language acquisition.

For children who are born severe-to-profoundly deaf it is known that age at implantation is one of the key factors that drives outcome (Sharma et al., 2004; Nicholas and Geers, 2010; Zhou et al 2013) but there is no clear consensus on all the prognostic indicators of outcome, or which individuals might be at higher risk of limited benefit postoperatively (Pisoni et al 2017). Other factors that are thought to be important are type of hearing loss (congenital onset of severe-to-profound deafness, progressive increase in hearing thresholds over time, acquired losses), degree of residual hearing pre-implantation and whether it is preserved, underlying etiology, presence of coexisting disabilities, socioeconomic status and consistency of use (Wiseman et al. 2021, Wiseman and Warner-Kzyz, 2018, Park et al. 2019). To fully understand the outcomes, we need to reflect upon the complex make up of patient groups and monitor how this has changed over the years in order to understand the extrinsic and intrinsic influences affecting outcomes. Blamey et al. (1996;2012) compared how the profile of factors affecting long-term outcomes in adult CI recipients changed over a 15 year period and showed that the importance of the different factors had changed. They attributed the differences in predictive profiles to relaxation of patient selection criteria, improved clinical management approaches, refinements in surgical practice, and improved devices.

In this research we have monitored the change in speech and language outcomes for children who received CIs over a 20-year period at a large inner-city hospital in the UK with the goal of understanding which factors affect long-term outcomes, how these factors have changed over time and how they were affected by changes in healthcare practice. Over the 20 year period, examples of changes in healthcare practice are the introduction of Universal Neonatal Hearing Screening in the UK which was phased in between 2002-2006 (Wood et al., 2015), the introduction of the UK National Institute of Health and Care Excellence (NICE) guidance on cochlear implants (TAG166, NICE 2009) which relaxed the indications for cochlear implants. This resulted in all children being offered bilateral rather than unilateral implantation as a standard of care. The UK ethnic and cultural diversity has also changed over the 20-year period due to migratory patterns within the population.

Traditionally, research has focused on children with congenital severe-to-profound deafness who were implanted in infancy or those whose hearing loss was progressive with a post-lingual (after 5 years) origin (Holt and Svirsky – 2008, Tajudeen et al – 2010, Wie 2020, Lovett et al. 2018). Researchers often exclude children for whom the native language was not their first or only language (Geers and Sedey 2010, Sharma 2017, Lovett et al. 2018). This is partly because many language outcome studies are directed at looking at effects relevant for the 'traditional' CI candidate.

For children growing up with a CI it is expected that they will become competent in the main language of the country in which they live. This is because education is provided in that language and if they can become proficient in that language it will most likely provide greater opportunities for further education and employment. Therefore, regardless of home language, clinical language assessments are typically undertaken in the main language spoken in that country. Exceptions would be for countries where a second language has significant community recognition and status (e.g. the USA (Spanish and English), Belgium (French and Flemish), South Africa (English and Africaans).

In this research we explored the 5 year post implant language scores for children provided with a CI between 1998-2019 to understand how the population had changed and what the profile of predictors of performance was when all children were included. In addition, we qualitatively compared the 5 year language outcomes from our research to those of a more traditional inclusion criteria range (e.g. based on Lovett, Vickers and Summerfield, 2018).

# Methods

The data collection was registered as a service evaluation with the University College London Hospital Audit Board

All children aged less than 18 years at the time of cochlear implant surgery were included. There were no exclusion criteria, randomisation or allocation to intervention groups, so no selection bias was introduced.

Retrospective data were collected from medical records for 418 children (total population) implanted between January 1998 and December 2019. Routine Speech and Language Therapy (SaLT) measures were conducted using the child's typical device configuration (bilateral cochlear implants or unilateral cochlear implant +/- hearing aid) and were collated. All assessments were conducted in a quiet clinical setting following administration guidelines. All SaLTs were specialists in working with deaf children and were experienced CI practitioners.

At each assessment interval the core language score (1-4) derived from the Clinical Evaluation of Language Fundamentals (CELF) was collected. The raw scores were converted to age-appropriate values to determine if language levels were 1, average or above; 2, mild or moderate delay; 3, severe delay; 4, not able to be tested.

The demographic data in this analysis included:

- age at implantation (months)
- implant status (unilateral/bimodal/bilateral simultaneous or sequential)
- type (onset) of hearing loss (congenital severe-to-profound, progressive, acquired)
- home languages (English 1st language (E1L), English additional language (EAL) & British Sign Language (BSL))
- postcode – as a socio-economic indicator. Postcode was converted to scores out of ten for the multiple indices of deprivation (based on national census database). An overall score and scores separated into seven categories were provided. The seven categories were parental education/skills, income, employment, health and disability, crime, barriers to housing, and living environment.

# Results

## Changes in predictors over time

The profile of change of patient characteristics over time was explored for home language distribution, type of hearing loss, socio-economic status, implant status and age at implantation.

To understand the pattern of changes over time six-year groups were created: 1998-2003, 2004-2006, 2007-2009, 2010-2012, 2013-2015, 2016-2019.

Home language distribution changed over the entire time period with 25% of children being from a family who did not speak English as their first language at home in 1998-2003 rising to 66% in the 2016-2019 time period (see figure 1). There was a significant change across the years for the distribution of children from native English-speaking families (E1L) and those from families where English is an additional language (EAL; $\chi 2=39.4$, df=5, $p<0.001$; BSL cases too low for inclusion). A significant jump in the distribution arose between the 3rd (2007-2009) and 4th (2010-2012) timepoints ($\chi 2=6.9$, df=1, $p=0.009$).

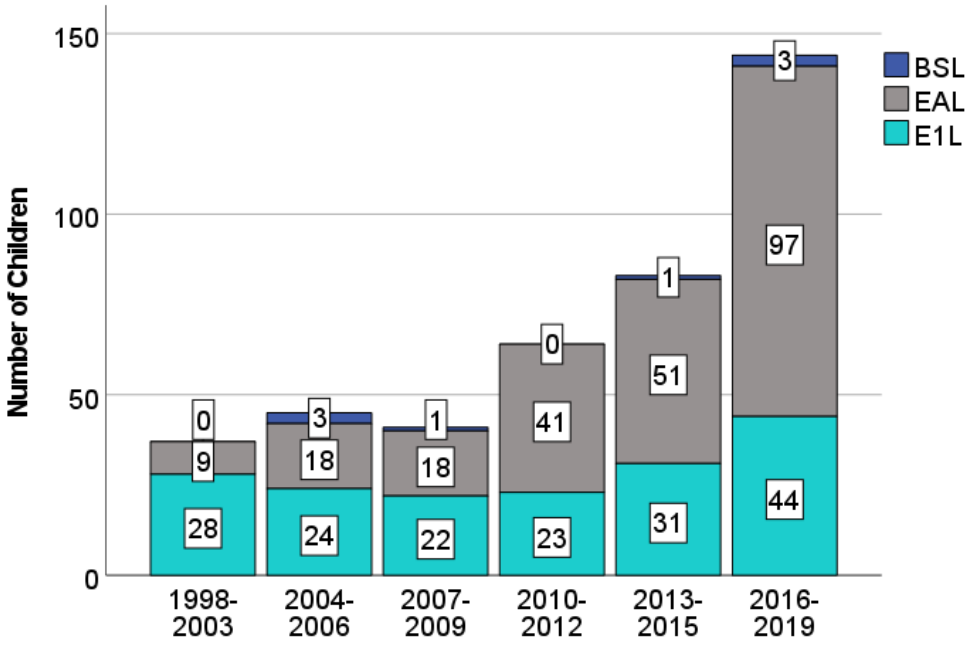


Figure 1 — Home language distribution across the 6 year groups. Number of cases shown in white boxes. BSL=British Sign Language, EAL=English as and additional language, E1L=English as a first language

There was a significant change in type of hearing loss (congenital severe-to-profound, progressive, acquired) across the years, ($\chi 2=32.2$, df=10, $p<0.001$). There was a marked increase in the proportion of children with progressive losses which was approximately 8% of the population in the earliest year group, rising to approximately 45% in the final year group (see figure 2).

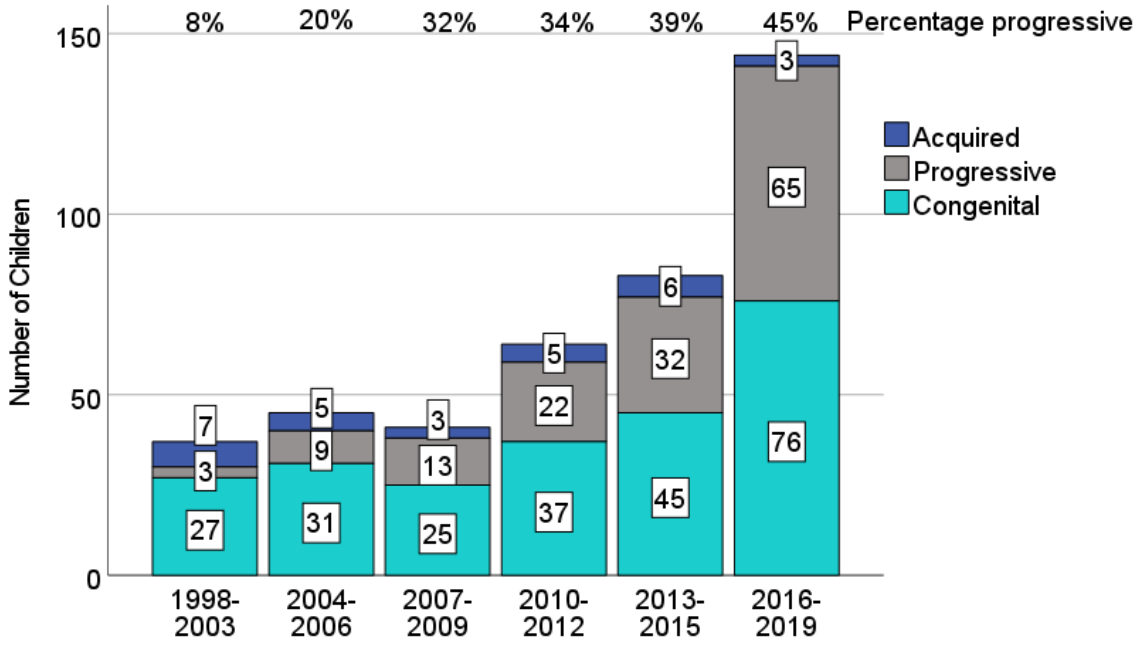


Figure 2 — Distribution of type of hearing loss over time as a function of year group. The percentage of progressives in each year group are indicated at the top. Numbers in white boxes are the absolute numbers in each sub category.

For socio-economic status the index of multiple deprivation was determined based on postcode using the national census for England figures. Percentile values for overall multiple deprivation showed a significant reduction (One-way ANOVA: $F(5,382)=3.47, df=5, p = 0.004$) across the year groups as shown in figure 3, where a lower score indicates greater deprivation. Post-hoc Tukey HSD tests ($p<0.05$ criteria) indicated that the last two year groups (2013-2015 and 2016-2019) were significantly lower than the first year group (1998-2003). The third group (2010-2012) showed a trend ($p=0.06$) towards being lower than the first year group.

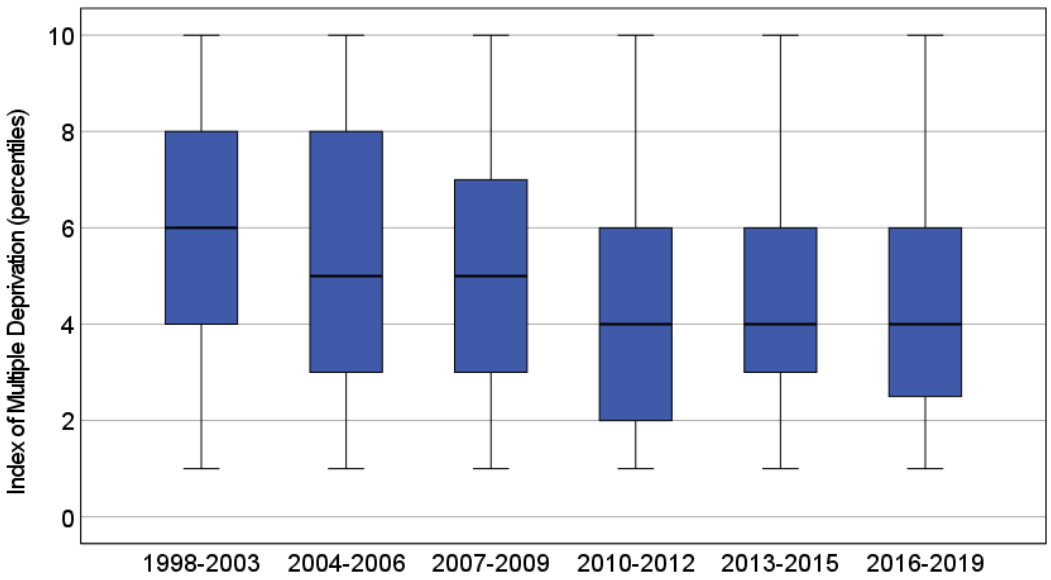


Figure 3 — Index of multiple deprivation (percentiles) over time. Lower score indicates greater deprivation

There were changes in implant status over time due to the introduction of the NICE guidance in 2019 which resulted in all appropriate children being given simultaneous bilateral CIs. In 2019 all children with unilateral implants were offered a sequential implant. This pattern can be observed in figure 4. The data are plotted according to the year of the first CI. The majority of children had bilateral implants.

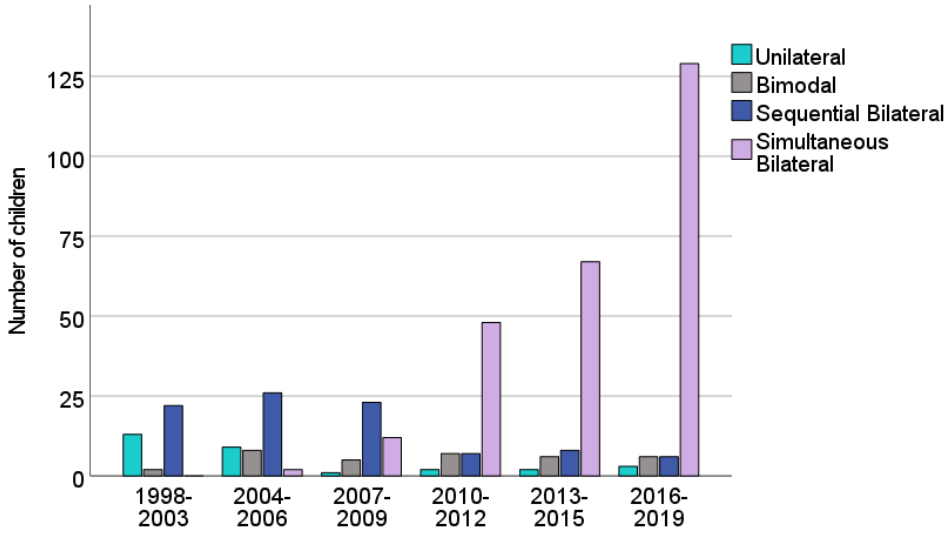


Figure 4 — Implant status as a function of year group.

For the analysis of age at implantation we only included children who were severe-to-profoundly deaf from birth (referred to as congenital; see figure 5). A Kruskal-Wallis test indicated that there was a significant change in age at implantation across the years ($\chi 2=21.12$, df=5, $p < 0.001$). There was a drop in median age across the first five age groups and at the later time point there was an increase in age. The early age drop is predicted to be due to the introduction of neonatal screening and the increase in the last group is most likely due to a change in candidacy criteria in 2019.

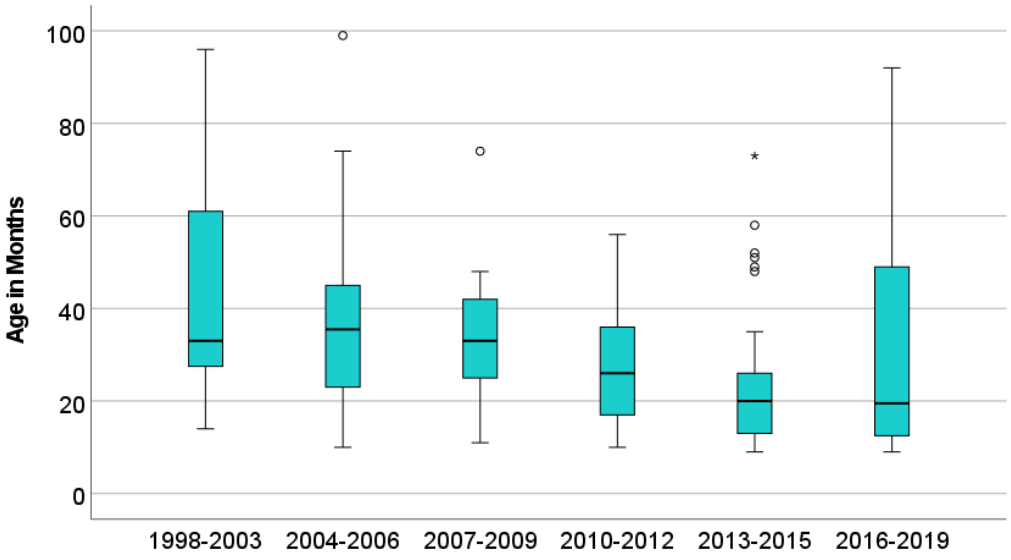


Figure 5 — Age at implantation as a function of year group over time.

## Predictive model of core language scores

A multinomial logistic regression was conducted to understand the factors influencing the core language scores (4 categories). The factors explored were *home language* (EAL, E1L), *type of loss* (congenital severe-to-profound, progressive, acquired), *age at implant* (months), *implant status* (unilateral, bimodal, sequential bilateral, simultaneous bilateral) and *multiple indices of deprivation*. For the multiple indices of deprivation the seven sub-categories were included separately as factors (*parental education/skills, income, employment, health and disability, crime, barriers to housing, and living environment*).

The fit between the model containing only the intercept and core language scores improved with the addition of the predictor variables *onset, home language, implant status and parental education and skills*, ($\chi 2$(df=21, N = 169) = 67.8, Nagelkerke R2 = .37, $p < .001$).

## Comparison of language scores with traditional experimental inclusion criteria

An exploratory comparison between core language outcomes from this research using all children who had reached the 5 year post implant stage and scores for a subset of children who fell within standard experimental inclusion criteria is shown in figure 6. The 'strict' restrictions are (1) Deafness identified before 31 months, (2) bilateral implant recipients, (3) native English speaking family. Using the strict inclusion criteria the findings indicated that 56% of the population had a moderate delay or better. Without any restrictions on inclusion the percentage with a moderate delay or better was 35%. With the restricted criteria it reduced the population to 25% of the unrestricted population.

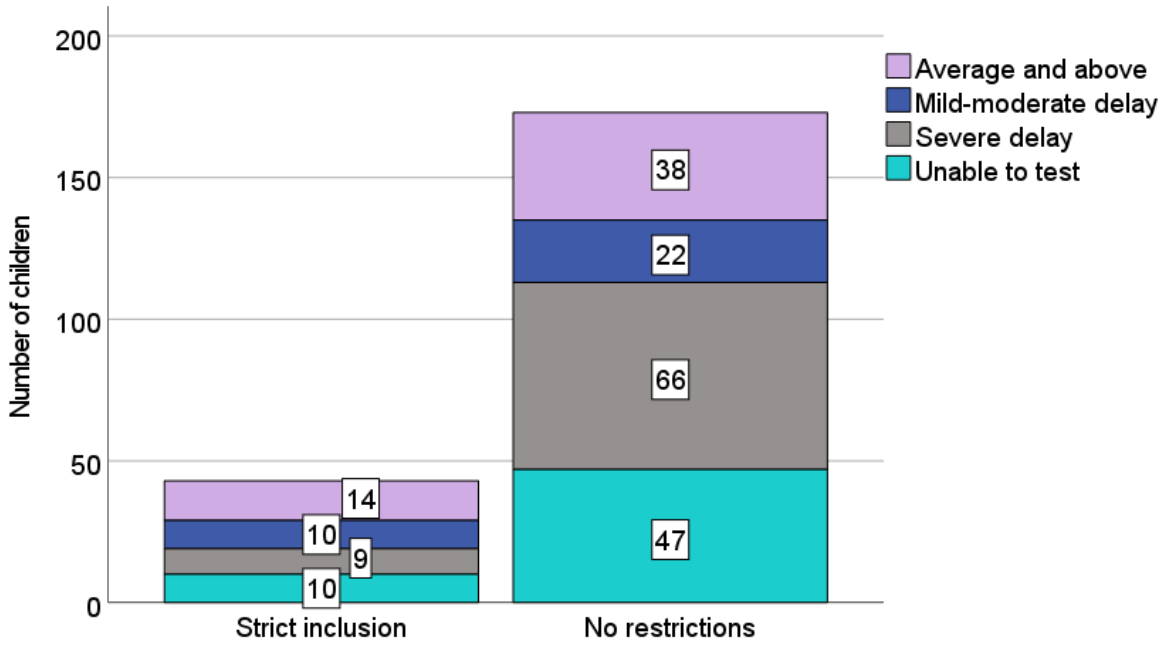


Figure 6 — The distribution of results according to inclusion criteria. Strict inclusion criteria based on typical publications and No restrictions indicates the data in this research.

# Discussion

Many of the factors that changed significantly over time (home language, type of loss, socio-economic status (specifically parental education level) and implant status) affected the language outcomes of children with CIs. Age at implant did not show as an indicator of outcome but this finding should be treated cautiously because our analysis included different types of hearing loss

onset (not purely congenital severe-to-profound onset). Age at implant dropped following the introduction of neonatal screening and continued to drop due to awareness of the importance of implanting early. However, in 2019, there was the introduction of an amendment to the NICE guidance on cochlear implantation and this resulted in individuals who had previously fallen outside criteria being implanted and they were generally older than the typical implant candidate at that stage. In future analyses etiology will be explored, to understand how it has changed over the years and the predictive contribution to outcome that it has. The introduction of genome mapping has led to better patient profiling. As there are changes in the ethnicity of the population there is also a chance that this affects the distribution of etiology.

Bilateral implantation was associated with better outcomes, however the children who did not have bilateral implants tended to have complex needs and therefore had decided not to have a second implant, so this may also be a finding to be treated cautiously.

For children whose home language was not English, they potentially could have had good language skills in the families native language but were poor in measures of English language. Anecdotally, this was not what was being reported by families but this is an area that future research should explore. It is however necessary to be able to fully engage in society to have a good grasp of the language where the child is being educated.

The overall finding is that long-term speech and language outcomes for the majority of children with CIs is poor. Only 35% of the population had age appropriate (or mild-moderate delay) language at 5 years post implantation. The largest area for focusing clinical efforts is that of improving family language skills.

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# References

*Being finalised*