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1	TITLE PAGE
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3	Factors Affecting Sound-Source Localization in Children with
4	Simultaneous or Sequential Bilateral Cochlear Implants.
5	
6	Catherine Killan ¹ , Andrew Scally ² , Edward Killan ³ , Catherine Totten ¹ and
7	Christopher Raine ¹
8	
9	¹ Yorkshire Auditory Implant Service, Bradford Royal Infirmary, Bradford, United Kingdom.
10	² School of Allied Health Professions and Sport, University of Bradford, Bradford, United Kingdom.
11	³ School of Medicine, University of Leeds, Leeds, United Kingdom.
12	
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20	Address correspondence to: Mrs Catherine Killan, Yorkshire Auditory Implant Service, Listening
21	for Life Centre, Bradford Royal Infirmary, Duckworth Lane, Bradford, BD9 6RJ, United Kingdom.
22	Email: catherine.killan@bthft.nhs.uk
23	

ABSTRACT

25

26 **Objectives:**

The study aimed to determine the effect of inter-implant interval and onset of profound
deafness on sound localization in children with bilateral cochlear implants, controlling for
cochlear implant manufacturer, age and time since second implant.

30 **Design:**

31 The authors conducted a retrospective, observational study using routinely collected clinical 32 data. Participants were 127 bilaterally implanted children aged 4 years or older, tested at least 12 months post-second implant. Children used implants made by one of three manufacturers. 33 34 Sixty-five children were simultaneously implanted, of whom 43% were congenitally, 35 bilaterally profoundly deaf at 2 and 4 kHz and 57% had acquired or progressive hearing loss. 36 Sixty-two were implanted sequentially (median inter-implant interval = 58 months, range 3 to 37 143 months) of whom 77% had congenital and 23% acquired or progressive bilateral 38 profound deafness at 2 and 4 kHz. Children participated in a sound-source localization test with stimuli presented in a random order from 5 loudspeakers at -60, -30, 0, +30 and +60 39 degrees azimuth. Stimuli were pre-recorded female voices at randomly roved levels from 65 40 to 75 dB(A). Root mean square (RMS) errors were calculated. Localization data were 41 42 analysed via multivariable linear regression models, one applied to the whole group and the 43 other to just the simultaneously implanted children.

44 **Results:**

45 Mean RMS error was 25.4 degrees (SD = 12.5 degrees) with results ranging from perfect 46 accuracy to chance level (0 to 62.7 degrees RMS error). Compared to simultaneous 47 implantation, an inter-implant interval was associated with worse localization by 1.7 degrees 48 RMS error per year (p < 0.001). Compared to congenital deafness, each year with hearing

thresholds better than 90 dB HL at 2 and 4 kHz bilaterally prior to implantation led to more accurate localization by 1.3 degrees RMS error (p < 0.005). Every year post-second implant led to better accuracy by 1.6 degrees RMS error (p < 0.05). Med-El was associated with more accurate localization than Cochlear by 5.8 degrees RMS error (p < 0.01) and with more accurate localization than Advanced Bionics by 9.2 degrees RMS error (p < 0.05).

54 Conclusions:

Inter-implant interval and congenital profound hearing loss both led to worse accuracy in sound-source localization for children using bilateral cochlear implants. Inter-implant delay should therefore be minimized for children with bilateral profound hearing loss. Children presenting with acquired or progressive hearing loss can be expected to localize better via bilateral cochlear implants than their congenitally deaf peers.

INTRODUCTION

62

Spatial listening includes the ability to hear where sounds come from. This skill is important 63 64 for children in social, recreational and educational settings as well as for their personal safety. 65 In individuals with normal hearing, sound localization (i.e. the ability to indicate which of multiple sound-sources a sound came from, in the horizontal plane) is possible because 66 central auditory processing makes use of inter-aural level differences (ILDs), inter-aural time 67 68 differences (ITDs) and spectral cues (Musicant & Butler, 1984). These cues are disrupted for 69 people with hearing impairment (Noble et al., 1994). One aim of auditory rehabilitation is to 70 restore binaural cues by providing appropriate hearing technology for both ears, with the 71 hope that spatial listening skills might be restored or developed to some extent despite the 72 hearing impairment.

73

74 Profoundly deaf children can learn to localize sounds significantly better with two cochlear 75 implants (CIs) than one (Steffens et al. 2008; Lovett et al. 2010; Godar & Litovsky 2010; Galvin et al. 2010; Grieco Calub & Litovsky 2010; Vincent et al. 2012). However 76 localization accuracy for children with bilateral CIs (BiCIs) varies from near-normal to an 77 inability to localize above chance level (Grieco-Calub & Litovsky 2010; Van Deun et al. 78 79 2010, Murphy et al. 2011). The reasons for this variation appear complex and are not yet 80 fully understood (Litovsky & Gordon 2016). It is likely that clinicians and CI manufacturers can influence some of the factors affecting sound localization, therefore greater 81 82 understanding of this area could benefit many profoundly deaf children.

83

A number of variables with the potential to influence localization have been identified by
previous researchers. First, auditory deprivation is likely to negatively affect localization via

86 neural degeneration and cortical reorganization (Sharma et al. 2007a, 2007b; Sparreboom et al. 2011; Gordon et al. 2011; Litovsky & Gordon 2016). Consistent with this, shorter inter-87 implant interval, younger age at second implant (CI2) and later onset of hearing-impairment 88 89 are associated with better localization in behavioural studies of children (Steffens et al. 2008; Grieco-Calub & Litovsky 2010; Van Deun et al 2010; Strom-Roum et al. 2012; Asp et al. 90 91 2015). However, a limitation of these studies is the high inter-correlation of time-dependent variables, e.g. age at first CI (CI1), age at CI2, age at test and inter-implant interval (Grieco-92 93 Calub & Litovsky 2010; Van Deun et al., 2010; Asp et al. 2015) which makes differentiation 94 between the effects of these variables difficult. Further, due to other limitations in previous studies, the effect of inter-implant interval on localization ability is not well understood. For 95 example, most studies are likely underpowered due to insufficient sample size (Van Deun et 96 97 al., 2010; Asp et al., 2011; Vincent et al., 2012), or make comparisons across different populations, i.e. post-lingually deafened adults versus congenitally deaf children (Litovsky et 98 al., 2004). 99

100

101 A second factor with the potential to influence localization ability is the cochlear implant 102 system used. Physical characteristics such as the number, separation and insertion depth of 103 the electrodes vary between systems, as do microphone characteristics. Each of these could feasibly influence the binaural perception of sound. Differences in speech processing 104 105 strategies including peak-picking versus continuous interleaved stimulation (Wilson et al., 1991), and variation in the knee-points, speed, complexity and frequency specificity of 106 compression circuits may be influential on ILD representation (Vaerenberg et al., 2014). 107 108 Representations of temporal fine structure also differ between systems and might feasibly 109 influence a user's ability to exploit ITDs (Eklöf & Tideholm, 2018; Thakkar et al., 2018). It is

therefore important to control for differences in implant and speech processing characteristicswhen examining localization outcomes.

112

Finally, age and binaural listening experience are known to influence children's localization outcomes. Normally-hearing children continue to improve on tests of sound localization until the age of around 5 to 7 years (Van Deun et al. 2009; Lovett et al. 2012). For sequentially implanted children, localization skills develop over the course of several years after receiving their CI2 (Litovsky et al. 2006; Asp et al. 2011; Kühn et al. 2013; Sparreboom et al. 2015). It is therefore important that age-related changes are accounted for when investigating localization ability development in CI users.

120

Given these potential influences on localization ability, and our lack of understanding of how they impact sound-source localization, the present study aimed to investigate the effects of inter-implant interval and onset of profound deafness on sound-source localization, whilst controlling for CI manufacturer, age, and time since CI2 on both simultaneously and sequentially implanted children.

127

METHODS

128

129 Participant selection and data collection

130 This study was a retrospective, observational study using routinely collected clinical data. Children using BiCIs, under the care of our service, aged 4 years or older and without 131 132 language and/or developmental delay that would preclude participation (as assessed by a relevant professional at a prior clinical appointment) were invited for assessment. In line 133 134 with candidacy criteria in the UK (NICE TAG 166, 2009) unaided hearing threshold levels 135 were 90 dB HL or worse at 2 and 4 kHz bilaterally prior to implantation. Data were excluded 136 from analysis for children with visual impairment that prevented them from seeing the 137 loudspeakers (N=1), children who withdrew co-operation before completing the full number 138 of test trials (N=1), children whose language comprehension was not sufficient to understand 139 the task (N=3), and children who completed the test trials but were uncooperative or 140 distracted to the point where the tester deemed their responses unreliable (N=7). Sound-141 source localization data from the remaining 127 children, with interval post-CI2 ranging from 1 to 6 years, were analysed. Characteristics of these children are summarised in Table 1. 142

143

144 Measurement of sound-source localization

Tests were administered via the A-B-York Crescent of Sound (Kitterick et al. 2011), a semicircular array of loudspeakers and monitors. The loudspeakers (Plus XS.2., Canton) were arranged at a height of 1.1m in a semi-circle of radius 1.45m and were controlled by custom software that produced simultaneous output via a digital-to-analogue converter (Ultralite Mk3, MOTU) and five dual-channel amplifiers (RA-150, Alesis). The software also controlled video monitors situated below the active loudspeakers, used as part of the patient response. The children sat on a chair in front of a table, facing the central loudspeaker and

152 equidistant from all loudspeakers. The study used the localization test developed by Kitterick et al (2011) and Lovett et al. (2012). Active loudspeaker locations were located at -60, -30, 153 154 0, +30 and +60 degrees azimuth (negative angles denote locations to the left). The positions 155 of the test equipment relative to the child are shown in Figure 1. The test software dictated that the maximum number of active loudspeakers was five. Stimuli were pre-recorded voices 156 157 saying "Hello, what's this?". Five different female talkers were used, 1 of whom was randomly selected on each trial by the software. The average presentation level was 70 158 dB(A), randomly roved by ± 5 dB in 1 dB steps. Children were instructed to face directly 159 160 ahead whilst listening for the stimuli, however no attempts were made to restrict head-161 movements during stimulus presentation.

162

163 The assessments were usually administered by one audiologist working alone. This tester would be seated across the room, operating the equipment via a desk-top keyboard and 164 165 monitor. A second tester was also present if this was recommended in the child's medical 166 notes. The second tester sat close by the child to help focus their attention. The testers were not blinded to the child's medical history or implant model as knowledge of these was 167 necessary to provide informed clinical care, ensure device function and counsel the family. 168 However, as this was a retrospective study, testers were blind to how the data would be used 169 170 for the purposes of this study.

171

One training presentation was given from each of the 5 active loudspeakers and for these the children were shown which loudspeaker the voice had come from. Every child then proceeded to the test trials regardless of their accuracy on the training trials. Six test stimuli were presented from each active loudspeaker so that there were 30 test trials. The test software randomly varied the loudspeaker from which stimuli were presented. Two methods

177 could be used for the localization test, to suit each child's interest and ability. The video monitors could show numbers 1 to 5 beneath each active loudspeaker and the child was asked 178 179 to say the corresponding number or to point to the loudspeaker that they thought the sound 180 came from. The alternative method involved placing coloured blocks of differing shape in front of the child whilst each monitor displayed a photograph of a different block. The child's 181 182 task was to locate the source of the sound and pick up the block displayed on the monitor below that loudspeaker. Children's continuing participation was praised regardless of their 183 184 accuracy. Reminders to listen were given as needed.

185

186 Analysis

187 For each child, sound-source localization accuracy was measured via RMS error of the 30 188 test trials. Linear multivariable regression models were used to explore the effects of a 189 number of explanatory variables on sound-source localization accuracy. These were inter-190 implant interval, age at onset of profound deafness, CI manufacturer, chronological age and 191 time post-CI2. Inter-implant interval, age at onset of profound deafness, chronological age 192 and time post-CI2 were continuous variables measured in months. Profound hearing loss was defined as unaided hearing threshold levels of 90 dB HL or worse at 2 and 4 kHz (these 193 194 frequencies are used to determine candidacy in the UK and hearing threshold levels at other frequencies therefore were not always available). CI manufacturer was entered into the model 195 196 as a categorical variable with Med-El arbitrarily chosen as the reference category. For each child left and right CIs were from the same manufacturer. Speech processor models were 197 always the same for right and left ears, however CI electrode array model sometimes differed 198 199 between ears, e.g. if a newer system was available at the time a second, sequential CI was 200 given. In one case a simultaneously implanted child had been re-implanted with a different 201 model following unilateral device failure (See Table 1). Regardless of manufacturer, all

children were programmed in omnidirectional microphone mode. See Table 2 for a summary
of CI system characteristics. As shown in Table 1, children using Advanced Bionics devices
were all simultaneously implanted. To determine whether this limited our analysis, the effect
of CI manufacturer on sound-source localization accuracy was also explored via a regression
model using data from simultaneously implanted children only.

207

208 For congenitally deaf children, older age at CI1 and CI2 imply longer periods of auditory 209 deprivation. This can be detrimental to sound localization ability (Van Deun et al. 2010; 210 Grieco-Calub et al. 2010). However with acquired and progressive losses, older age at CI1 211 and CI2 may be due to having useful acoustic hearing for a longer time, resulting in less 212 auditory deprivation, which might be expected to improve a child's localization with CIs 213 (Grieco-Calub & Litovsky 2010; Killan et al. 2015). For these reasons, whilst age at CI1 and 214 CI2 were recorded, they were not used in the regression analyses. Table 3 shows ages at CI1 215 and CI2 by manufacturer for the children born with profound hearing loss at 2 and 4 kHz in at 216 least one ear.

RESULTS

219

220 Across all 127 children RMS error ranged from perfect accuracy to chance, i.e. 0.0 to 62.7 degrees¹. Mean RMS error was 25.4 degrees (standard deviation, SD = 12.5 degrees). Figure 221 2 shows a scatterplot of RMS error as a function of inter-implant interval (months). Data-222 223 points at 0 months are from children who received simultaneous CIs. The mean RMS error 224 for this group was 21.6 degrees (SD = 11.07 degrees), with a range broadly consistent with 225 that seen for the remaining sequentially implanted children (mean RMS error = 29.5 degrees; 226 SD = 12.7 degrees). RMS error is seen to increase with increasing inter-implant interval. Table 4 shows the results of the regression model, which confirms this relationship. Each 227 228 month's wait between CI1 and CI2 was associated with less accurate localization by 0.14 229 degrees RMS error (p < 0.001).

230

231 Figure 3 shows the relationship between RMS error and age at onset of profound hearing loss 232 (months). In this figure the data-points at 0 months are from congenitally profoundly deaf children. This group had mean RMS error of 28.6 degrees (SD = 12.1 degrees) and a similar 233 range of RMS error to the acquired/progressive children, i.e. the other data-points in the 234 figure, whose mean RMS error was 20.7 degrees (SD = 10.5 degrees). RMS error is seen to 235 decrease with increasing age at onset of hearing loss. This was shown to be a significant 236 237 effect by regression analysis, with RMS error improving by 0.11 degrees for each month 238 delay in the onset of bilateral profound hearing loss (p < 0.01).

239

RMS error is plotted against age at test in Figure 4. No clear trend is evident, as confirmed by regression analysis (p = 0.47). Figure 5 shows RMS error against time post-CI2 (months).

¹Chance performance being 62 degrees RMS error, Pádraig Kitterick, personal communication.

242 Data points are clustered around 1, 2 and 4 years post-CI2, as these were standard assessment 243 intervals. Although trends are difficult to discern from subjective inspection of the figure regression analysis, controlling for the other variables, found a significant reduction of 0.13 244 degrees per month post-CI2 (p < 0.05). Significant differences were obtained between the 245 246 manufacturers. Med-El systems were associated with more accurate localization, with RMS error 5.79 degrees smaller than Cochlear (p < 0.01) and 9.19 degrees smaller than Advanced 247 248 Bionics (p < 0.05). As all Advanced Bionics users were simultaneously-implanted, a second model exploring the effect of CI manufacturer using only data from simultaneously implanted 249 250 children was performed. This gave similar results to the first model, suggesting that the 251 differences in localization seen between CI systems was not materially affected by whether a 252 child received their implant simultaneously or sequentially (Table 5).

254 **DISCUSSION**

255

256 Previous research is limited in its ability to provide information on potential influencing 257 factors on sound-source localization ability. Limitations include small sample size, inability to differentiate between the effects of time-based factors and comparisons across different 258 259 populations. The present study therefore aimed to investigate the effects of factors that 260 influence localization ability, namely inter-implant interval and onset of profound deafness, 261 whilst controlling for CI manufacturer, age and time since CI2, for simultaneously and 262 sequentially implanted children. This was achieved via multiple regression analysis of 263 routinely collected clinical data from a large number of children. This allowed analysis of the 264 independent effects of a number of explanatory variables on localization accuracy (measured 265 via RMS error).

266

We found a broad range of localization accuracy, consistent with other studies of bilaterally 267 268 implanted children. Our whole group mean RMS error was 25.4 degrees, ranging from 0.0 to 269 62.7 degrees. Zheng et al (2015) reported mean RMS errors of 19 children, 4 of whom had some acoustic experience prior to BiCI and 8 of whom had less than one year inter-implant 270 271 interval. When first assessed with mean BiCI experience of 29.8 months, mean RMS error was 31.3 degrees, falling to 26.2 degrees for the same children at a later assessment interval. 272 273 This is comparable to the present study. Grieco-Calub and Litovsky (2010) report mean RMS 274 error of 37.4 degrees (range 19 to 56 degrees) for 19 sequentially implanted children, around 275 half of whom were congenitally deaf. Van Deun et al. (2010) report a very similar mean RMS 276 error of 38 degrees from 30 children who were all implanted sequentially and two thirds of 277 whom were congenitally deaf. Compared to Grieco-Calub & Litovsky (2010) and Van Deun 278 et al. (2010) our children localized with smaller RMS error on average. This may be due to the relatively longer duration of BiCI experience of the children in our study, the larger proportions of children with acquired and progressive losses and simultaneous implantation, and likely methodological differences also. Asp et al. (2011) reported bilaterally implanted children's localization in terms of Error Index. Outcomes also varied from perfect accuracy to chance performance for a five loudspeaker array localization task. We found greater mean RMS error than that of normally-hearing children, who typically perform the task with perfect accuracy (Lovett et al. 2012).

286

287 Inter-implant interval was shown to have a significant influence on sound-source localization 288 ability. This adds to the arguments in support of minimizing inter-implant interval where 289 possible for children with bilateral profound hearing loss. It is interesting to note that the 290 effect of inter-implant interval was still significant in this group who had received their 291 second implant up to 6 years ago. Thus, despite being experienced users of bilateral CIs, children did not fully overcome the detriment caused by prolonged inter-implant interval. 292 293 This is consistent with theories of long-lasting cortical reorganization in response to unilateral 294 auditory deprivation, which suggest a critical period of 18 months (e.g. Gordon et al. 2013; 2015). 295

296

Our data showed age at onset of bilateral profound hearing loss also had a significant effect on sound-source localization, with better performance seen for children with longer experience of bilateral acoustic sound prior to BiCIs. This is consistent with previous studies that indirectly explored the effect of auditory experience during the early years. For example, Grieco-Calub & Litovsky (2010) showed that children reported by parents to be benefiting from hearing aids were more likely to have better sound-source localization via CIs than peers who had not benefitted from hearing aid use. Previously, Killan et al. (2015) showed

that type of hearing loss (categorized as acquired/progressive or congenital) influenced sound-source localization, with children with acquired/progressive loss performing better than those with congenital hearing loss. Their study was limited due to children with acquired/progressive loss being older than children in the congenitally deaf group. The present data adds to the evidence for the effect of age at onset of profound hearing loss by quantifying and directly exploring this variable.

310

311 All children whose unaided hearing thresholds were outside NICE CI criteria (NICE, 2009) 312 up to at least approximately 48 months of age localized with better accuracy than the group 313 average in the present study. This is broadly consistent with reports by Sharma et al., (2007a, 314 2007b) who showed that congenitally deaf children need to receive CIs in both ears by the 315 age of 42 months to give symmetrical electrophysiological responses to sound. Other 316 behavioural studies of localization ability in children have also noted a benefit of binaural 317 listening during the early years (Steffens et al., 2008; Grieco-Calub & Litovsky, 2010; Van 318 Deun et al., 2010). It should be noted however that in the present study, pre-operative 319 hearing thresholds below 2 kHz were not used in our definition of profound hearing loss. As 320 a consequence it is possible that the present study may underestimate the effect of pre-321 operative hearing levels due to unknown variability in low-frequency hearing.

322

323 CI manufacturer had a significant effect on sound-source localization ability, with Med-El 324 implants being associated with the most accurate localization ability, followed by Cochlear 325 and then Advanced Bionics. Interpretation of this effect based on modelling the whole group 326 was potentially complicated since all users of Advanced Bionics devices were simultaneously 327 implanted, whilst Med-El and Cochlear users were either simultaneously or sequentially 328 implanted. It was therefore possible that the effect of manufacturer seen might have been 329 influenced by inter-implant interval effects. To explore this, an additional model including 330 data from only simultaneously implanted children (regardless of CI manufacturer) was 331 calculated and compared with the original model. Similar differences between the three 332 manufacturers were evident in both models, indicating that CI manufacturer had a similar effect on localization accuracy for both simultaneously and sequentially implanted children 333 334 and that this effect was independent of inter-implant interval. Our study was not designed to explore reasons for differences between systems however, one plausible explanation may be 335 differences in automatic gain control, which can influence outcomes for speech 336 337 discrimination (Spahr et al. 2007). The range of stimulus intensity used in the present study, 338 from 65 to 75 dB(A), is toward the higher levels for speech, equivalent to e.g. a team-mate 339 calling during a sports game or raised voices during a group meal. Input sounds from 65 to 75 340 dB SPL may result in CI stimulation at levels at, or close to, maximum stimulation amplitude 341 for Cochlear and Advanced Bionics patients, hence ILDs for sounds in this range may be 342 difficult to perceive. For Med-El recipients these intensities are mapped to a lower portion of 343 the patients' dynamic range and will produce CI stimulation over a wider range of electrical 344 amplitude (Vaerenberg et al. 2014). Localization via CIs is thought to be dominated by ILDs (Seeber & Fastl 2008), even in children where fine structure strategies facilitate some degree 345 346 of ITD sensitivity (Eklöf & Tideholm, 2018). Since Med-El systems apply less compression 347 to sounds louder than 65 dB SPL compared to Cochlear or Advanced Bionics, ILD cues may 348 have been better preserved for children using Med-El systems in the present study. It is therefore possible that repeating this study using quieter stimuli would not find the same 349 350 difference between manufacturers. Age at CI1 and CI2 is not likely to account for the 351 difference in localization across CI manufacturers. On average, for congenitally deaf 352 children, Med-El users were older at CI1 and CI2. This is a detrimental influence on sound-353 source localization, not advantageous, and so cannot explain the results.

354

Consistent with previous studies (Asp et al., 2011; Kühn et al. 2013; Asp et al. 2015), time 355 356 post-CI2 was shown to influence localization ability, with a longer time associated with 357 improved performance. It should be noted, however, that actual binaural listening time varies from child to child dependent on how consistently they use their devices. Inconsistent device 358 359 use has been shown to be a particular issue for sequentially implanted children (Galvin & Hughes. 2012; Fitzgerald et al. 2013). A limitation of the present study was that it was not 360 361 possible to determine or control for how much time each child had spent listening via both 362 CIs together.

363

364 One limitation of this study is the use of RMS error alone to measure sound localization, as it 365 does not capture more subtle aspects of a person's localization ability (Grieco-Calub & Litovsky, 2010; Zheng et al., 2015; Killan et al., 2018). A further potential limitation is the 30 366 degrees spacing between loudspeakers, which does not allow localization accuracy to be 367 368 measured with the fine spatial resolution achieved in some other studies (e.g. Zheng et al., 2015). However, as the mean RMS error found in the present study (25.4 degrees) is 369 comparable to those reported by Zheng et al. (2015) (31.3 and 26.2 degrees at first and 370 371 second test intervals respectively) it is likely that loudspeaker separation did not substantially 372 impact our findings. Indeed, the spatial resolution achieved in our study is similar to other 373 previous research (e.g. Asp et al., 2011; Killan et al., 2015; Murphy et al., 2011). A loudspeaker array with large separations between speakers may lead to ceiling effects, where 374 children find the test too easy. As only one out of the 127 children completed the test with 375 376 perfect accuracy, it is considered unlikely that ceiling effects limited our findings. Similarly, 377 Asp et al (2011) reported only two out of sixty-six bilaterally implanted children perfectly completed a localization task that used five loudspeakers separated by 45 degrees. 378

379 Importantly, the loudspeaker spacing in the present study is representative of situations a 380 hearing impaired child might encounter in day to day life, at mealtimes, during lessons or 381 playing sport. For example, a child might be writing while sat around a table with friends 382 doing group work at school when another child begins to speak.

383

384 Our regression model accounted for 26% of variance in the data. A number of factors not measured in our study potentially account for some of the remaining variance. One such 385 factor is asymmetric loudness growth caused by, for example, avoidance of facial nerve 386 387 stimulation or recent re-programming. A second possible influence is the effect of children moving their head following stimulus onset. Whilst asked to face ahead for the onset of each 388 389 presentation, some children moved their heads more than others during the sentence and 390 some leaned forward when they were concentrating, effectively moving the loudspeaker array 391 out of the horizontal plane. Finally, variation in children's concentration during the task could 392 account for some variance in the data. Data were excluded from analysis if the tester deemed 393 a child had been uncooperative or distracted such that their responses were clearly unreliable. 394 However the analysed group will have included children with varying levels of attention, 395 fatigue and motivation, potentially influencing their responses in more subtle ways that are 396 difficult to quantify.

397

Our findings provide further evidence that the unilateral auditory deprivation experienced while waiting between a first and second CI causes a long-term detriment in subsequent sound-source localization. Therefore inter-implant interval should be minimized for children with bilateral severe-to-profound hearing loss. The significant effect of age at onset of profound deafness means that clinicians can expect children with acquired or progressive hearing loss to localize comparatively well via BiCIs. This knowledge is useful for patient

404 selection for BiCIs, counselling and targeting rehabilitation for children where progress is not 405 seen. Another implication is that it is important to monitor a child's localization accuracy over several years following BiCIs so that failure to develop localization skills can be 406 407 identified. Where this is found, appropriate measures can be taken to initiate targeted 408 rehabilitation, including reviewing BiCI use, addressing any programming issues, or 409 recommending localization listening practice. The family can be counselled regarding their 410 child's speech processor use to ensure that both processors are worn simultaneously for most of the day, rather than alternating, and also to ensure that processor microphones are not 411 412 positioned side-by-side on the top of the child's head rather than over the ears. Listening 413 practice can include games where the child closes their eyes and family members play an 414 instrument from differing, unknown locations in the room, then the child guesses where the 415 sound came from; or where a noise-making toy or phone is hidden in the room and the child is encouraged to listen to help them find it. 416

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431	C.F.K. contributed to the conception and design of the work, collected the data, contributed
432	to the interpretation of data analysis, drafted the article, contributed to the article's revision
433	and gave final approval of the submission. A.J.S. conducted the data analyses, contributed to
434	the writing of the results section of the manuscript, and gave final approval of the submission.
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438	the submission. C.H.R. contributed to the conception of the work, revision of the draft article
439	and gave final approval of the submission.
440	
441	Address for Correspondence:

442 Mrs Catherine Killan

- 443 Clinical Scientist (Audiology)
- 444 Yorkshire Auditory Implant Service
- 445 Listening for Life Centre
- 446 Bradford Royal Infirmary
- 447 Duckworth Lane
- 448 BD9 6RJ
- 449 Email: catherine.killan@bthft.nhs.uk

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594 Figure 1

595 The child is shown seated in front of a table and facing the centre of the arc of 596 loudspeakers. Loudspeaker positions are shown in degrees azimuth, negative angles 597 denote locations to the left and positive angles denote locations to the right of centre. 598 Inactive video monitors and loudspeakers are shown in grey. Active video monitors and 599 loudspeakers are shown in black.





Figure 2

RMS errors are plotted for each child against the duration of their inter-implantinterval in months.



609 Figure 3

610 RMS errors are plotted for each child against the age at which their hearing loss was 611 first confirmed to be profound at 2 and 4 kHz in at least one ear. Children whose 612 hearing impairment was detected by newborn hearing screening and confirmed to fall 613 within this range on immediate follow-up are plotted as having met this criterion from 614 birth and are clustered at the far left.

615



618 Figure 4

RMS errors are plotted for each child against their age at test in months.



623 Figure 5

624 **RMS** errors are plotted for each child against the number of months since they received

625 their second CI.

627 Table 1628 Participant Characteristics

		Simultaneous	Sequential
		(N = 65)	(N = 62)
Age (months)	Median	91	136
	Youngest	52	85
	Oldest	235	307
Onset of profound hearing	Congenital	N = 28 (43%)	N = 48 (77%)
loss categorized	Acquired / Progressive	N = 37 (57%)	N = 14 (23%)
Age at onset of profound	Median	10	0
deafness at 2kHz and 4 kHz	Youngest	0	0
in at least one ear (months)	Oldest	185	68
Age at CI1 (months)	Median	50	31
	Youngest	6	14
	Oldest	220	165
Age at CI2 (months)	Median	50	88
	Youngest	6	26
	Oldest	220	283
Inter-implant Interval	Median	0	58
(months)	Least	0	3
	Greatest	0	143
Time since CI2 (months)	Median	47	48
	Least	12	13
	Greatest	77	84
Manufacturer	Med-El	N = 27 (42%)	N = 38 (61%)
	Cochlear	N = 31 (48%)	N = 24 (39%)
	Advanced Bionics	N = 7 (11%)	N = 0 (0%)
Children with differing Cl models in right and left ears		N = 1 (1%)	N = 48 (77%)

632 Table 2 ristia 633 634 child f th .,, chla Ch 4 ar imnlant ct.

Characteristics (of the children	's cochlear	implant systems

Advanced Bionics (N=7)	Cochlear (N=55)	Med-El (N=65)
Naida (N=2)	CP910 (N=5)	Opus 2
Harmony (N=5)	CP810 (N=52)	
	Freedom (N=1)	
HiRes Optima S (N=2) HiRes-S w/Fidelity 120 (N=5)	ACE	FSP
Processor, omnidirectional	Standard, omnidirectional	Standard, omnidirectiona
Up to 16	Up to 22	Up to 12
15 mm	15 to 20 mm	23 to 26 mm
	Advanced Bionics (N=7) Naida (N=2) Harmony (N=5) HiRes Optima S (N=2) HiRes-S w/Fidelity 120 (N=5) Processor, omnidirectional Up to 16 15 mm	Advanced Bionics (N=7)Cochlear (N=5)Naida (N=2)CP910 (N=5)Harmony (N=5)CP810 (N=52)Freedom (N=1)Freedom (N=1)HiRes Optima S (N=2)ACEHiRes-S w/Fidelity 120 (N=5)ACEProcessor, omnidirectionalStandard, omnidirectionalUp to 16Up to 2215 mm15 to 20 mm

Table 3Ages at CI1 and CI2 by manufacturer for the congenitally deaf children

	Age at CI1 (months)		Age at CI2 (months)	
	Median (range)		Median	(range)
Advanced Bionics (N=4)	20	(13 to 67)	20	(13 to 67)
Cochlear (N=29)	26	(12 to 186)	63	(12 to 186)
Med-El (N=43)	32	(6 to 216)	91	(6 to 216)
	52	(0 10 210)	51	(0

642 **Table 4**

- 643 Results of regression analysis for both simultaneously and sequentially implanted
- 644 children.

645

No = 127

No = 127 Adj. R ² = 0.259					
Variable		Coefficient	p	95% Cor Inte	nfidence rval
Inter-Implant Interval (mont	hs)	0.14	<0.001	0.07	0.22
Onset of Deafness: Age when HTLs ≥90 dB HL at 2 and 4 kHz in at least one ear first measured (months)		-0.11	0.004	-0.19	-0.04
Time since CI2 (months)		-0.13	0.035	-0.26	-0.01
Age at Test (per month of life)		-0.02	0.466	-0.07	0.03
Manufacturer (relative to Med-El)	Cochlear	5.79	0.006	1.65	9.93
(,	Advanced Bionics	9.19	0.043	0.31	18.06

646

647 A positive coefficient indicates an association between the variable and greater, i.e. less

648 accurate SLA.

649

Table 5

Results of regression analysis for simultaneously implanted children only.

No = 65 Adj. R ² = 0.220					
Variable		Coefficient	р	95% Cor Inte	nfidence rval
Onset of Deafness: Age when HTLs ≥90 dB HL a least one ear first measured	at 2 and 4 kHz in at (months)	-0.10	0.007	-0.17	-0.03
Time since CI2 (months)		-0.17	0.033	-0.33	-0.01
Age at Test (Per month of lif	e)	-0.03	0.213	-0.09	0.02
Manufacturer (relative to Med-el)	Cochlear	7.48	0.008	2.03	12.93
(Advanced Bionics	10.66	0.017	1.97	19.36

655 A positive coefficient indicates an association between the variable and greater, i.e. less

accurate SLA.