

The impact of hearing loss on speech outcomes in 5-year-old children with Cleft Palate ± Lip: A longitudinal cohort study

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ABSTRACT

Objectives

To investigate the impact of hearing loss (using longitudinal measurements of hearing) on speech outcomes at age 5 (5 years 0 months- 5 years 11 months) in children born with cleft palate ± lip. Other variables which may impact upon the speech outcomes at age 5 in this population were also investigated.

Methods

A retrospective longitudinal cohort study of children, without a named syndrome, born with cleft palate ± lip, and treated at a Cleft Centre in the United Kingdom. Data collected from infancy to 5 years 11 months, included hearing test results from three specific time points (7 months – 1 year 2 months [age A]; 2 years 0 months – 2 years 11 months [age B]; 5 years 0 months – 5 years 11 months [age C]) and speech outcome data at age 5 years (5 years 0 months – 5 years 11 months). Hearing test results at each age were compared to identify how hearing changes with age. Correlations between hearing test results and speech outcomes at age 5 were analysed.

Results

Hearing loss was frequent but predominantly mild. There were no significant correlations between speech outcomes and hearing results at any age. Mild hearing loss remained prevalent at age 5, although a significant age-related hearing improvement was found. A significant relationship between cleft type and cleft speech characteristics was found ($P < .001$); children with Bilateral Cleft Lip and Palate achieved the poorest articulation outcomes.

Conclusion

Although mild hearing loss was common in the cohort, there was no association between hearing loss and the speech outcomes investigated. In contrast, the type of cleft was significantly associated with the presence of cleft speech characteristics. Further longitudinal measurement of hearing is required to substantiate the findings of this study.

1. Introduction

Children with cleft palate \pm lip (CP \pm L) are at risk of conductive hearing loss secondary to Otitis Media with Effusion (OME) [1, 2] which is almost 'universal' in this population. Children with CP \pm L are also an 'at risk' group for speech impairment due to the structural and functional impact of the palatal cleft [3]. The Cleft Care UK review [4, 5] of speech outcomes for children with Unilateral Cleft Lip and Palate highlighted that just under 20% of the participants had the lowest scores for intelligibility/distinctiveness at age 5 (5 years 0 months-5 years 11 months). To improve speech outcomes, it is important to identify groups within the CP \pm L population who may be most at risk for a poor speech outcome at age 5. Children with Robin Sequence [6], and children with Bilateral Cleft Lip and Palate (BCLP) who have undergone lip adhesion [7] have been identified as potential 'at risk' groups for poor speech outcomes at age 5. Given the prevalence of conductive hearing loss in this population [1, 8] it is important to consider what effect this may have on speech development.

The literature investigating the impact of hearing on speech outcomes in children with CP \pm L is inconclusive. Some authors [9-14] have identified a significant link between hearing and speech outcomes. This includes Lohmander et al. (2011) [14] who reported an association between hearing at 12 months of age which was not seen at 18 months. Others have found no association [14-17] between hearing and speech. Such variation in findings could be accounted for by methodological differences resulting in heterogeneous studies, with notable differences in the study cohorts, and the measures of hearing and speech used. For example, the age range of participants in the studies differs significantly, ranging from 12 months [14] to 20 years [17]. There are also notable differences in the parameters of speech assessed in the studies. For example, Magnus et al. (2011) [12] and Hall et al. (2017) [13] assessed speech

intelligibility (referring to the proportion of which speech sounds are correctly identified and understood), both reporting a significant link between hearing and speech. In contrast, Schönweiler et al. (1999) [9] reported a significant link between hearing and phonology outcomes whilst Klintö et al. (2014) [15], also reporting on phonology outcomes, did not. The impact of hearing on cleft specific articulation outcomes also varies [10-11, 15-17]. The reliability of those studies who used 'live' listener judgements of speech and did not report on listener reliability should also be questioned [9-11, 17].

There is significant variation in how hearing is assessed and reported in previous studies. Firstly, hearing thresholds varied widely, with hearing loss measured as >16dB [17], whilst in another study >27dB [12]. The validity of those studies measuring hearing at a single time point, in a population at risk for fluctuating hearing loss which changes over time [18], should also be questioned [9-10, 12, 16-17]. The studies by Lohmander et al. (2011) and Klintö et al. (2014) [14, 15] demonstrate the importance of measuring hearing over time. The two studies follow the same group of children, reporting an association between hearing and speech outcomes at 12 months, but not at 18 months or 3 years. The validity of proxy measures used to measure hearing impairment should similarly be questioned. Hall et al. (2017) used a history of ventilation tube (VT) insertion or hearing aid use as an indication of hearing impairment. This is problematic given these measures do not directly relate to hearing levels; VT can be inserted for reasons other than to treat hearing loss, and because insertion rates of VT tubes are known to vary between Cleft Centres [19]. The number of participants in previous studies also varies widely from 18 children with CP±L [15] to 370 [12] which poses challenges when interpreting results across the studies.

This study aimed to address those methodological concerns of previous studies. Hearing was assessed at three-time points at a single Cleft Centre with lower-than-average ventilation tube (VT) insertion rates [19]. A reliable and valid speech assessment [20, 21] designed specifically to measure those outcomes key to the assessment of cleft speech [22] was utilised, this included the assessment of nasality outcomes given that previous studies have reported significant associations between hearing and this parameter [10-11]. Secondary objectives focused on assessing longitudinal changes in hearing and identifying other clinical outcomes which may impact speech outcomes at age 5.

2. Material and Methods

2.1 Ethical Approval

Ethical Approval was gained from Coventry University and Birmingham Women's and Children's NHS Trust (P40263)

2.2 Study Design

A quantitative study utilising retrospective analysis of prospectively collected hearing and speech outcome data in a cohort of children with CP±L. Data were prospectively collected between 2005 and 2014 and retrospectively analysed between 2016-2017. Patient and Public Involvement (PPI) was central to the development of the project and PPI groups were involved at both the educational and hospital institutions.

2.3 Participants

Participants were recruited from a large Cleft Centre in the United Kingdom. All children had their cleft repair carried out by one of three surgeons at the Cleft Centre. A

sample size of 85 was determined using Cohen's method [23], assuming a p-value of $p = 0.05$ and a medium effect size.

Children were included if they were born between 2005-2009, had complete longitudinal hearing data and complete speech outcome data at age 5 (5 years 0 months- 5 years 11 months). These dates were selected as the children born in 2005 were the first cohort for which validated speech outcomes were available at age 5, and those born in 2009 were those for whom the most recent speech outcome data at age 5 was available. Longitudinal hearing data referred to hearing test results within three age bands: 7 months- 1 year 2 months (age A), 2 years 0 months – 2 years 11 months (age B) and 5 years 0 months – 5 years 11 months (age C).

Children were excluded if they had a diagnosed syndrome, if their speech assessment results at age 5 were incomplete, if their longitudinal hearing assessment results were incomplete at any of the above-mentioned age bands, or if the child was treated with hearing aids (indicating a sensorineural hearing loss) during this period. Children with named syndromes were excluded from the study, as speech outcomes for this group are not reported as standard at age 5 in the UK [24] and because speech outcomes for children with named syndromes and cleft palate can differ from the wider cleft population [25].

Between 2005 and 2009, 428 children were born with CP±L and treated at the Cleft Centre. Of these children, 262 had complete speech assessment results at age 5. Of these, 150 were excluded due to incomplete longitudinal hearing data. A further 24 children having been treated with hearing aids were excluded, leaving a sample of $n=88$.

Participant gender, Cleft Type, cleft surgeon (one of three surgeons), and any history of VT insertions was recorded.

2.4 Audiology Assessment

Visual Reinforcement Audiometry (VRA) or Pure Tone Audiometry (PTA) were performed by trained paediatric audiologists. VRA was carried out in free field and is not ear specific. If there were concerns regarding sound localisation, ear inserts were utilised. PTA was carried out using headphones.

A hearing frequency average for each hearing test result was calculated; for ear-specific testing, the frequency average of the better hearing ear was used. If at any age-point the child did not complete four-frequency testing (500Hz, 1kHz, 2kHz and 4kHz), an average of the available frequencies was used. The frequency average was then classified per the current British Society of Audiology (BSA) guidelines [26, 27]. For PTA the classification was: normal hearing: ≤ 19 dB; mild hearing loss: 20-40 dB; moderate hearing loss: 41-70 dB; severe hearing loss: 71-95 dB; profound hearing loss: ≥ 96 dB. For tests carried out in the free-field, each classification was increased by 10dB.

2.5 Speech Assessment

All children had a Cleft Audit Protocol for Speech-Augmented (CAPS-A) [20] score at age 5. This outcome is used to measure speech outcomes at age 5 in all UK Cleft Centres. The CAPS-A is a valid and reliable speech outcome measure [20, 21] when used by Speech and Language Therapists (SLTs) trained to use the tool. The CAPS-A provides an overall score in two parameters: i. Nasality and ii. Cleft Speech Characteristics (CSCs: patterns of speech associated with CP±L). These parameters are core to the assessment of cleft speech [22] and

are measured on a traffic light score (dark green, light green, amber and red). The optimum score is a 'green' score in both areas (dark or light green) indicating that the cleft has had minimal impact on the individual's speech outcome. Within the parameters of Nasality and CSCs there are different subgroups. The child's overall score for Nasality and CSCs is formed by taking their worst score from each subgroup. Nasality is rated in four different subgroups (hypernasality, hyponasality, nasal emission and nasal turbulence) with a traffic light scale for each. For CSCs, four main classes of CSC are rated (anterior-oral CSCs, posterior-oral CSCs, non-oral CSCs and passive CSCs). Each of these classes contains further subgroups referring to specific types of CSC, all scored on a traffic light system. Specific descriptors for each subgroup are used to determine which traffic light score is most appropriate. Developmental speech sound substitutions (not cleft related) are classified as present or absent.

All of the children had a speech assessment comprised of a spontaneous speech sample, counting 1-20, counting 60-70, reciting a nursery rhyme and repeating sentences taken from the Great Ormond Street Speech Assessment [28] which was video recorded. Following the CAPS-A protocol, a process of consensus listening was used in which 2+ specialist cleft SLTs trained to use the CAPS-A tool, watched the recordings, and rated the children's speech. The protocol requires a minimum of 10% of Cleft Centre speech assessments to be rated by SLTs external to the Cleft Centre.

2.7 Statistical Analysis

Statistical Analysis was carried out using IBM SPSS version 24 and Microsoft Excel 2016. Descriptive statistics were used to describe the demographics of the cohort and both hearing test and speech outcomes. Inferential statistics were used to calculate correlation (parametric data: Pearson's Correlation coefficient; Non-parametric data: Spearman's Correlation

coefficient), association (Chi-square test or Fishers exact), variance (one-way repeated measures ANOVA) and to compare means (one-sample *t*-test, paired-sample *t*-test, independent *t*-test). A significance level of $p < .05$ was used.

3. Results

3.1 Demographic Information

Of the cohort, $n=52$ (59%) had cleft palate (CP) only, $n= 26$ (30%) had unilateral cleft lip and palate (UCLP) and $n=10$ (11%) had bilateral cleft lip and palate (BCLP); $n=43$ (49%) of the children were female and $n=45$ (51%) male. When grouped according to surgeons who had performed cleft repair surgery, the following split was noted (surgeon 1: 40%, surgeon 2: 37% and surgeon 3: 23%).

3.2 Hearing Outcomes

All children had VRA testing at age A and age B and PTA testing at age C. Within the cohort, three categories of hearing were identified (normal hearing, mild hearing loss and moderate hearing loss). No children were identified with severe or profound hearing loss. Most children (82%) completed an assessment at four or more frequencies at age C, compared to 39% at age A. All participants had ear specific testing at age C. 41% had better hearing in the left ear, 32% had better hearing in the right ear, and 27% had similar hearing in both ears. For age A, 10% had normal hearing, 83% had mild hearing loss and 7% had moderate hearing loss. At age B, 40% had normal hearing, 56% had mild hearing loss and 4% moderate hearing loss. At age C, 50% had normal hearing and the rest had mild hearing loss.

Although mild hearing loss was common at age C, there was a statistically significant improvement in hearing with age; mean frequency average at age A was 26.07(\pm 9.24)dB, age B 23.11(\pm 9.13)dB and age C 18.16(\pm 7.05)dB. A one-way repeated measures ANOVA to compare the frequency average at each hearing test point, indicated significant differences across age groups ($F(2.0, 174) = 21.219, P = .001, \eta p^2 = .196$). Post hoc tests using Bonferroni correction ($0.5 / 3 = .16$) indicated statistically significant frequency average improvement with age for all comparisons (age A-age B: $t = 2.250; P = .027, d = .322$; age A-age C: $t = 6.721; P = .001, d = .962$; age B-age C: $t = 4.183; P = .001, d = .607$). Hearing improved at each frequency assessed.

Normal hearing outcomes at all the three age bands were found in only 3% of children, 25% had two normal tests. Of the cohort, 32% had hearing loss at each test point. Using Spearman's correlation coefficient, there was no significant correlation between cleft type and the hearing frequency average score at any of the three assessment points (age A $P = .518$; age B $P = .177$; age C $P = .645$).

Of the cohort, $n = 26$ (30%) children had VT inserted during the study period. The VT of choice was a Shah grommet, no other type of VT was used during the study period. Most children had VT inserted for the first time between their second and third birthday ($n = 8, 31\%$). The children's age at initial VT insertion is presented in Table 1, with a range of 20-68 months. Of the 26 children who underwent VT insertion, 11 (42%) had more than one set of VT inserted within the study period, with a range of 1-3 VT insertions. The indication for VT insertion in 24 children (92.3%) was a parental concern about hearing and a hearing loss on audiometry; 3 children suffered from recurrent acute otitis media; and 1 child had retraction of the tympanic membrane. Audiometry results prior to VT insertion were available for 25

children (these results may or may not have been one of the 3 audiometry time points used in this study). 1 child did not condition for testing but as there was parental concern VT were inserted. Prior to VT insertion, 80% of children (n=20) had mild loss, and 20% (n=5) had moderate loss. Children with VT had a worse frequency average hearing score than children without VT, however independent *t*-tests indicate that this difference was only significant at age B, as outlined in Table 2.

<Insert Table 1 about here>

<Insert Table 2 about here >

3.3 Speech Outcomes

To determine whether the speech outcomes of the study sample (n=88) were representative of those of the Cleft Centre, a one sample *t*-test was used. CAPS-A results for Nasality and CSCs for all children born between 2005-2009 (n=262) were calculated; no significant differences of the study cohort (n=88) were found ($t = -.875, P = .422$), suggesting that the study sample findings are representative of the CAPS-A results for all children of the Cleft Centre.

A total of 66% (n=58) of the study sample achieved a green outcome (dark or light) for both Nasality and CSCs. 89% (n=78) had a green outcome (dark or light) for Nasality and 74% (n=65) had a green outcome (dark or light) for CSCs (Figure 1). An amber or red outcome for both CSCs and Nasality was found for 3% of the sample. Developmental errors were present in (n=48) 57% of the children, but data was missing for 3.

<Insert Figure 1 about here>

3.4 Correlation between Hearing and Speech Outcomes

Spearman's correlation coefficient indicated no significant correlation between the speech outcomes of CSCs or Nasality and the hearing frequency average at any of the hearing test points (Table 3). Fisher's exact test showed no association between the total number of normal hearing tests and the speech outcome for CSCs or Nasality (CSCs $P = .378$; Nasality $P = 7.86$).

<Insert Table 3 about here>

Spearman's rank correlation coefficients revealed no relationships between frequency average and any of the CSC categories or specific types of CSCs (backing to velar/uvular and active nasal fricatives) (Table 4).

<Insert Table 4 about here>

3.5 Other clinical outcomes and the Speech Outcome

Fisher's exact test was used to calculate the association between other clinical outcomes and CSCs or Nasality. A significant association between the CSC result and cleft type ($P = .001$) was found. Children with BCLP had the poorest CSC results, followed by UCLP and CP (Figure 2). The most common CSC in the BCLP group was posterior oral CSCs (60%) with 50% having evidence of backing to velar or uvular places of articulation. In contrast, there was no significant association between Nasality and cleft type ($P = .855$).

<Insert Figure 2 here>

Fisher's exact test revealed no significant association between CSCs and surgeon ($P = .311$) nor between Nasality and surgeon ($P = .959$), suggesting comparable surgical outcomes

across surgeons. There was no significant association between speech outcomes and VT insertion: CSCs ($P = 1.00$) and Nasality ($P = .274$).

4. Discussion

This study investigated the impact of longitudinal hearing on speech outcomes at age 5. This is important for two reasons, firstly hearing is known to both fluctuate and improve with age in this population [1] and this is substantiated by the statistically significant improvement in hearing observed in this study. Secondly, speech development from infancy to 5 years is a dynamic, developing, process and, as illustrated by Lohmander et al. [14], there may be different relationships between hearing and speech outcomes at different ages.

A limitation of previous studies investigating hearing and speech outcomes the CP±L population is the use of a single hearing test result [9-10, 12, 16-17] (which may not be representative of hearing at an earlier age), or the use of proxy measures such as VT insertion or hearing aid use to identify patients with persistent hearing loss [13]. Children requiring hearing aids cannot be directly compared to those needing VT insertion, as the former may have permanent mixed or sensorineural hearing loss where the latter may have mild to moderate temporary loss. Additionally, hearing loss is not the only indication for VT insertion [29]. As such, these proxy measures can be problematic, introducing a selection bias given that patients without VT or hearing aids may also suffer from hearing loss. Indeed, in the sample of the current study, the majority of children demonstrated some form of hearing loss at some point in time, with only 3% of the cohort demonstrating 'normal' hearing at all three test points. This suggests that studies exploring hearing outcomes should incorporate longitudinal hearing test data to limit the risk of error and bias, which may occur when proxy measures are used. Indeed, in this study, the VT insertion rate (30%) was much lower than

previously reported across other UK cleft centres [19], and none of the children in the sample had VTs inserted before 7 months of age. However, those with VT insertion were not found to have better speech outcomes, when compared to the group which did not require VTs.

No correlation was found between hearing and any of the speech outcomes investigated. In addition, children with a higher number of normal hearing test results did not achieve a statistically better outcome for CSCs or Nasality. In spoken English, the majority of speech sounds can still be perceived with a mild hearing loss [30] and the fluctuating nature of this loss, with the dynamic nature of speech produced at differing intensities and frequencies may also facilitate speech acquisition. This may suggest that for the CP±L population, mild hearing loss has limited functional impacts on CSCs. However, this finding is not congruent with some of the previous studies in this area [9-14]. A strict inclusion and exclusion criteria was adopted in this study in order to control for variables which may impact upon the speech and hearing outcomes. However, previous studies have included individuals with unrepaired palates [12], submucous cleft palates [9], individuals with cleft lip and alveolus only [9, 12], and individuals with syndromes [16], all of which may impact both hearing and speech outcomes. Differences in the way in which hearing was measured between previous studies may also account for the differing outcomes.

The CAPS-A [20] is cleft specific, measuring those speech outcomes most likely to be impacted by having a cleft. It is perhaps unsurprising given the good Nasality outcomes for the cohort that there was no association between this outcome and hearing. Whilst hypernasality is a feature in the speech of profoundly deaf individuals [31, 32] only mild and moderate hearing loss was seen in this study. It is challenging to fully account for the findings of other studies reporting a link between hypernasality and hearing [10-11]. The CAPS-A does

not, however, provide a consonant inventory and future research could consider if specific sounds are particularly vulnerable to hearing loss in the CP±L population. Magnus et al. [12] and Hall et al. [13] found a significant link between hearing and measures of intelligibility. Intelligibility is no longer part of the routine CAPS-A audit assessment [20] and is not referred to in the UK National Speech Standards for CP±L [24], due to concerns regarding the validity of this measure of speech.

Whilst hearing was not identified as a risk factor for speech outcomes at age 5, a significant association between cleft type and CSCs was found. Children with BCLP had the poorest speech outcomes (red or amber), whilst children with CP had the highest number of green outcomes. In contrast, there was no significant association between BCLP and Nasality outcomes. As all 3 surgeons operate at a high-volume Centre and have comparable experience, this may account for the lack of association between the surgeon and speech outcomes [33]. These findings may suggest that children with BCLP are at an at-risk group for poorer CSC outcomes, with wider implications for monitoring, service provision and service delivery.

This study faces several limitations. Given the retrospective analysis, full hearing data sets were only available for 88 out of 262 children with speech outcome data. Tympanometry results were incomplete and had to be excluded, which meant it was not possible to confirm the presence or absence of OME at each hearing test point. Whilst a single frequency (6% at age A and 10% at age B) hearing test result would have informed clinical decisions, it may not reflect hearing at other frequencies. Previous studies [9-13, 15-17] have assessed both speech and hearing outcomes at the same time. Although hearing was assessed longitudinally, speech was only assessed at a single time point. This raises questions regarding whether an

earlier association between hearing and speech may have resolved by the age of 5. We have no data on the frequency and duration of speech therapy interventions each child may have received and thus it is not possible to determine the impact of therapy on speech outcomes and if it mitigated the effect of hearing loss.

5. Conclusion

In this longitudinal study of 88 children with CP±L, hearing significantly improved over time, but mild hearing loss was still present in half of the participants at age 5. No correlations were found between the specific cleft speech outcomes and hearing results at any of the three assessment time points. Participants with BCLP had the poorest articulation outcomes and appeared to constitute a group 'at risk' of poor articulation outcomes. Further research measuring both hearing and speech longitudinally is required.

Highlights:

- Hearing significantly improves with age in the cleft palate ± lip population
- Mild hearing loss did not impact speech outcomes at age 5
- Children with Bilateral Cleft Lip and Palate have the poorest speech outcomes at age 5

Key words:

Cleft palate, cleft lip and palate, hearing, OME, speech, Nasality, Cleft Speech Characteristics

Table 1. Age at VT insertion.

Age at first VT (Shah grommet) insertion	Number of children (n=26)	Percentage of children with VT (%)
1 year 0 months- 1 year 11 months	4	15.4
2 years 0 months- 2 years 11 months	8	30.8
3 years 0 months- 3 years 11 months	6	23.0
4 years 0 months- 4 years 11 months	4	15.4
5 years 0 months-5 years 11 months	4	15.4

Table 2. Differences in Frequency Average between Children With and Without VT Insertion

Test age	Group	Mean	SD	<i>d</i>	<i>t</i>	<i>p</i>
Age A	VT	28.23	9.25	.332	-1.428	.157
	No VT	25.17	9.17			
Age B	VT	27.22	11.40	.605	-2.401	.020
	No VT	21.39	7.46			
Age C	VT	19.88	8.90	.320	-1.494	.105
	No VT	17.44	6.05			

Table 3. Correlations between Frequency Average and Speech Outcomes

Speech Outcome	Test Age	r_s	p
CSC	Age A	.187	.082
	Age B	-.099	.357
	Age C	.008	.939
Nasality	Age A	.039	.718
	Age B	.158	.142
	Age C	.067	.537
Overall result (CSC and nasality)	Age A	.037	.730
	Age B	.010	.923
	Age C	.046	.671
Developmental speech sound substitutions	Age A	.151	.160
	Age B	.051	.640
	Age C	-.074	.495

Table 4. Correlations between Frequency Average and Categories of CSCs/Specific Types of CSCs

Categories of CSCs/Specific Types of CSCs	Test Age	r_s	P
Anterior Oral CSCs	Age A	-.003	.980
	Age B	-.152	.157
	Age C	.047	.662
Posterior Oral CSCs	Age A	.204	.057
	Age B	-.084	.439
	Age C	-.025	.815
Non-Oral CSCs	Age A	.154	.152
	Age B	.079	.463
	Age C	.010	.924
Passive CSCs	Age A	-.029	.788
	Age B	.058	.595
	Age C	.073	.500
Backing	Age A	.156	.145
	Age B	-.063	.560
	Age C	-.057	.596
Active Nasal Fricatives	Age A	.124	.250
	Age B	.014	.896
	Age C	.019	.862

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