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

Beth Fitzpatrick*[a,b]. MScR

Channa Panagamuwa[a]. FRCS ORL-H&NS, MMedEd

Leora Moss-Levi[c]. PhD

Tanya Rihtman [b]. PhD

[a] Birmingham Children's Hospital, UK

[b] Coventry University, UK

[c] Kidma Centre, Israel

*Correspondence Address: Beth Fitzpatrick, Speech and Language Therapy Department, Birmingham Children's Hospital, Steelhouse Lane, Birmingham, B4 6NH, UK.

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ABSTRACT

Objectives

To investigate the impact of mild hearing loss on speech outcomes at age 5 years in children born with cleft palate ± lip. Secondary objectives were to measure hearing longitudinally from 7 months to 5 years and to identify other variables which may impact upon the speech outcomes at age 5 years.

Methods

A retrospective longitudinal cohort study of 88 children, born with cleft palate ± lip, and treated at a Cleft Centre in the United Kingdom. Data collected over 5 years included hearing test results from three-time points (7-14 months [age A]; 24-35 months [age B]; 60-71 months [age C]) and speech outcome data at age 5 years. Hearing test results at each age were compared to measure changes in hearing with age. Correlations between hearing test results and speech outcomes at age 5 were measured.

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 common in the cohort, there was no association between mild hearing loss and the speech outcomes investigated. This suggests that mild hearing loss does not disrupt the perception of speech sounds to the extent that it is a significant reason for Cleft Speech Characteristics or altered Nasality in children with cleft palate ± lip. On the other hand, 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 ± lip (CP±L) are at risk of conductive hearing loss secondary to Otitis Media with Effusion (OME) [1 Flynn & Lohmander, 2014, 2 Flynn et al. 2014] which is almost 'universal' in this population. Children with CP±L are also an 'at risk' group for speech impairment due to the structural and functional impact of the palatal cleft [3 Chapman et al. 2011]. The Cleft Care UK review [5 Sell et al. 2015, 6 Sell et al. 2017] 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. To improve speech outcomes, it is important to identify groups within the CP±L population who may be most at risk for a poor speech outcome at age 5 years. Children with Robin Sequence [7. Hardwicke et al.], and children with Bilateral Cleft Lip and Palate (BCLP) who have undergone lip adhesion [8. Peryer et al. inpress] have been identified as potential 'at risk' for poor speech outcomes at age 5. Given the prevalence of conductive hearing loss in this population [1 Flynn & Lohmander 2014 ,9 Imbery et al. 2017] it is important to consider whether hearing loss is a potential risk factor for speech development.

Whilst a significant link between hearing and speech outcomes has been reported in the CP±L population [10-15] this is not conclusive, and several other studies have not reported such an association [15-18]. Such differences in the results could be accounted for by the heterogeneity of the 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 [15] to 20 years [18]. There are also notable differences in the parameters of speech used in the studies. Magnus et al. (2011) and Hall et al. (2017) reported on the speech outcome of intelligibility, both reporting a

significant link between hearing and speech. In contrast, Schönweiler et al. (1999) and Klintö et al. (2014) both measured phonology outcomes yet reported different outcomes regarding the impact of hearing on phonology. The impact of hearing on cleft specific articulation outcomes also varies in the literature [11-12, 16-18].

Another notable difference within the studies is the method by which hearing was assessed, with some studies recording hearing loss at 16dB [18] and others from 27dB [13]. The majority of the studies tended to make use of single hearing test results [7-10] which may not represent the fluctuating nature of hearing loss due to OME, nor the potential changes in hearing over time [19 Gravel]. The benefit of testing hearing at more than one-time point is highlighted by Lohmander et al. (2011) and Klintö et al. (2014) [15, 16]. Lohmander et al. [15] reported an association between hearing and consonant inventory at age 12 months, which was not seen at 18 months of age. In a follow-up study by Klintö et al. [16], at age 3-years, differences in phonology outcomes persisted after hearing was been controlled for. Proxy measures, using a history of ventilation tube (VT) insertion or hearing aid use as a measure of hearing impairment [14] are problematic given that these measures do not directly relate to hearing levels, that 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 [20 Fitzsimmons].

The primary objective of this study was to determine the longitudinal impact of hearing on speech outcomes at age 5 years, using multiple hearing measures, at a single Cleft Centre with lower than average ventilation tube (VT) insertion rates [20]. Along with articulation outcomes, nasality outcomes were also investigated given that previous studies have reported significant associations between hearing and this parameter [11-12].

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 2009; data were retrospectively analyzed 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. Children were included if they were born between 2005-2009. These dates were selected as 2005 was the first year in which validated speech outcomes were available at the Cleft Centre, and 2009 constituted the most recent year for which longitudinal data sets of 5-year-old children were available. Longitudinal data sets referred to hearing test results within three age bands: 7-14 months (age A), 2 – 2 years and 11 months (age B) and 5 – 5 years and 11 months (age C).

The inclusion criteria stipulated speech outcome data at age 5 and the availability of longitudinal hearing data at all three ages. Children were excluded if they had a diagnosed syndrome in addition to CP±L, if their hearing assessment results were incomplete at any age point, if their speech assessment results at age 5 were incomplete, or if the child was treated with hearing aids during this period.

A sample size of 85 was determined using Cohen's method [21], assuming a p-value of $p = 0.05$ and a medium effect size. Between 2005 and 2009, 428 children were born with CP±L and treated at the Cleft Centre. Of these children, only 262 had complete speech assessment results at age 5. Of these, 150 were excluded due to incomplete hearing data. A further 24 children were excluded having been treated with hearing aids, leaving a sample of n=88.

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

2.4 Audiology Assessment

Visual Reinforcement Audiometry (VRA) or Pure Tone Audiometry (PTA) were administered at the Cleft Centre by trained audiologists. VRA was carried out in the 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 for that age. The frequency average was then

classified per the current British Society of Audiology (BSA) guidelines [22, 23]. 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: ≥ 96dB. 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) [24] score at age 5 years. This outcome is used to measure speech outcomes at age 5 years in all UK cleft centres. The CAPS-A is a valid and reliable speech outcome measure [24, 25 Sell et al. CAPSA paper] 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) 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). Within both 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 different traffic light scales. 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 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 [26] 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. In 41% better hearing was seen in the left ear, 32% had better hearing in the right ear, and 27% had the same result 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, ηp^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 of the three tests points were found for 3% of children, 25% had two normal tests and 40% had one normal test. 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 sample, n=26 (30%) children had VT inserted between 7 months and 5 years 11 months. Of these, most had VT inserted for the first time at age 2 years (n=8, 31%,). Children with VTs had a worse frequency average score than children without VTs, however independent *t*-tests indicate that this difference was only significant at age B, as outlined in Table 1.

<Insert Table 1 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 2). 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 2 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 3).

<Insert Table 3 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). 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. When assessing the impact of hearing on speech in the CP±L population, longitudinal hearing measurement is important given that hearing is known to both fluctuate and improve with age in this population [1, 27], and because Lohmander et al. [15] demonstrated that there may be differing relationships between hearing and speech outcomes at different ages. This is substantiated by the statistically significant improvement in hearing with age which was shown in this study. A limitation of previous studies investigating hearing and speech outcomes in this population is the use of a single hearing test results [10, 11, 13, 17, 18], or the use of proxy measures such as VT insertion or hearing aid use to identify patients with persistent hearing loss [14]. 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 (recurrent acute otitis media) [27 Wilson]. 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 ages. This suggests that studies exploring hearing outcomes should incorporate longitudinal hearing test data to ensure that no children are overlooked, which may occur when proxy measures are used. Given that

participants treated with hearing aids were excluded from the study, it is unsurprising that the most predominant type of hearing loss was mild, with moderate loss rarely seen.

An additional justification for the importance of exploring hearing outcomes relates to differences across centres in VT insertion rates. In the Cleft Centre in question, the VT insertion rate (30%) was much lower than previously reported across other UK cleft centres [20 Fitzsimons], and none of the children in the sample had VTs inserted before 7 months of age. This suggests that the Cleft Centre in question may be conservative with VT insertion. As such, it is likely that only children with persistent or severe symptoms would have had the procedure, and therefore may represent a subgroup within the cohort of children more likely to have hearing loss. However, no association was found between hearing and speech outcomes in the subgroup with VTs. This suggests that the insertion of VTs may potentially have supported more positive speech outcomes for this sub-group.

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 [20] 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 [10-15]. In this study a strict inclusion and exclusion criteria was used to control for variables which may impact upon the speech and hearing outcomes. However, previous studies have included individuals with unrepaired palates [13], submucus cleft palates [10], individuals with cleft

lip and alveolus only [10, 13], and individuals with syndromes [17], 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.

Both Magnus et al. [13] and Hall et al. [14] found a significant link between hearing and measures of intelligibility. Intelligibility was not measured in the present study, due to concerns regarding the validity of this measure of speech, it is no longer part of the routine CAPS-A audit assessment [24], and is not referred to in the UK National Speech Standards for CP±L [4 Britton]. The CAPS-A [24] whilst a valid and reliable measure of cleft speech, was not designed to assess speech impairment caused by hearing loss and it does not provide a consonant inventory. Future research could consider if specific sounds are particularly vulnerable to hearing loss in the CP±L population. 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 individual [28 Yunsa and Vasquez, 29 Kummer et al.] only mild and moderate hearing loss was seen in this study. Other studies reporing a link between hypernasality and hearing [11-12] may be reporting a link between the adequacy of palatal function and the subsequent impact on the eustachian tube function and in turn hearing, rather than a direct link between hearing and 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 [21]. 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 [10-14, 16-18] 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. All children in this study were regularly assessed by specialist SLTs and therapy sessions were provided to support speech development. As the study did not explore the nature of therapy provision, it is not possible to confirm whether this constitutes a confounding factor.

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 years. No correlations were found between speech outcomes (CSCs and Nasality) 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 years
- Children with Bilateral Cleft Lip and Palate have the poorest speech outcomes at age 5 years

Key words:

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

Figure 1. Frequency of Children achieving Dark Green, Light Green, Amber or Red Outcomes for CSC and Nasality

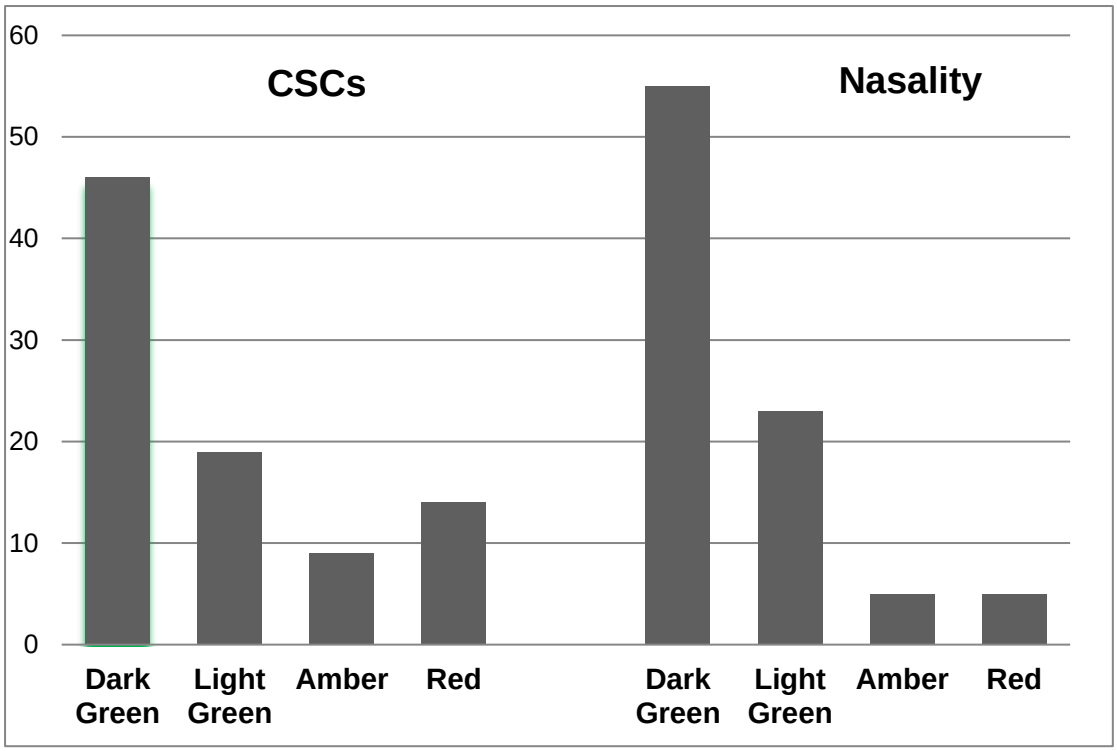

Figure 2. Traffic Light Category Result for CSCs by Cleft Type

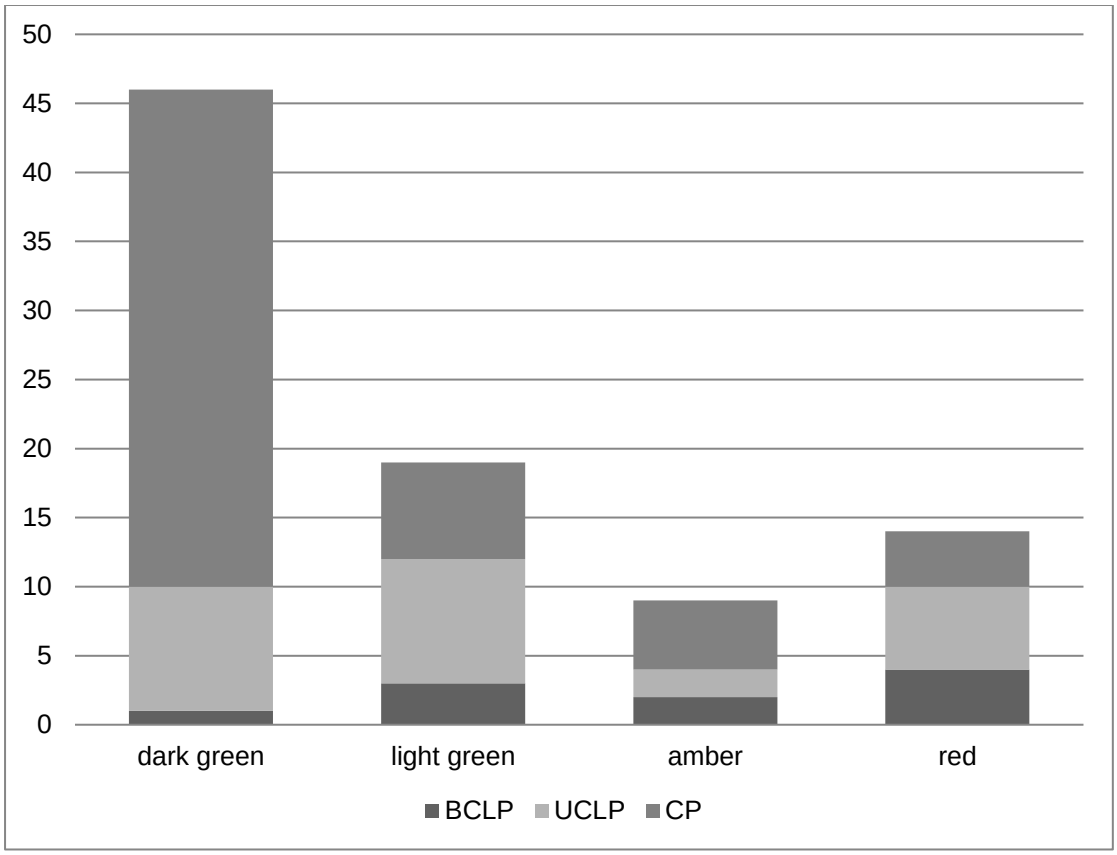

Table 1. Differences in Frequency Average between Patients With and Without VT Insertion

Test age	**Group**	**Mean**	**SD**	***d***	***t***	***p***
Age a	Grommets	28.23	9.25	.332	-1.428	.157
	No grommets	25.17	9.17			
Age b	Grommets	27.22	11.40	.605	-2.401	.020
	No grommets	21.39	7.46			
Age c	Grommets	19.88	8.90	.320	-1.494	.105
	No grommets	17.44	6.05			

Table 2. 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 3. 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