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ORIGINAL ARTICLE

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Centre-level variation in speech outcome and interventions, and factors associated with poor speech outcomes in 5-year-old children with non-syndromic unilateral cleft lip and palate: The Cleft Care UK study. Part 4

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Structured Abstract

Objectives: To investigate centre-level variation in speech intervention and outcome and factors associated with a speech disorder in children in Cleft Care UK (CCUK).

Setting and Sample Population: Two hundred and sixty-eight 5-year-old British children with non-syndromic unilateral cleft lip and palate recruited to CCUK.

Materials and Methods: Centre-based therapists undertook audio-video recordings. Perceptual analysis was undertaken using the CAPS-A tool. Speech outcomes were based on structural and articulation scores, and intelligibility/distinctiveness. Between-centre variation in treatment and outcomes were examined using multilevel models. These models were extended to estimate the association between a range of factors (hearing loss, speech intervention, fistula, secondary speech surgery for velopharyngeal insufficiency, socio-economic status, gender, and parental happiness with speech) and speech outcomes.

Results: There was centre-level variation in secondary speech surgery, speech intervention, structure and intelligibility outcomes. Children with a history of speech intervention had a lower odds of poor intelligibility/distinctiveness, 0.1 (95% CI: 0.0-0.4). Parental concern was associated with a higher odds of poor intelligibility/distinctiveness, 13.2 (95% CI: 4.9-35.1). Poor speech outcomes were associated with a fistula, secondary speech surgery and history of hearing loss.

Conclusions: Within the centralized service there is centre-level variation in secondary speech surgery, intervention and speech outcomes. These findings support the importance of early management of fistulae, effective management of velopharyngeal insufficiency and hearing impairment, and most importantly speech intervention in the preschool years. Parental concern about speech is a good indicator of speech status.

KEYWORDS
centralization, centre-level variation, cleft lip and palate, poor speech outcomes, secondary speech surgery, speech intervention

1 | INTRODUCTION

It has consistently been reported that there is a group of children and young people, born with cleft palate, who have long-term intractable speech impairment.[1,2] This is important as we know that maximizing speech skills in the preschool years is important for psychosocial well-being and educational reasons. Children with persisting speech concerns at age 5 years are more likely to present with later educational difficulties and long-term socio-economic consequences.[3–10] Identifying the factors associated with poor speech outcomes at 5 years of age may help us to design and target interventions and resources that improve this important outcome.

Studies of children born with unilateral cleft lip and palate (UCLP) have reported that up to 40% of Swedish children have ongoing articulation difficulties and/ or velopharyngeal dysfunction at the age of 5 years.[11–13] Hypernasality and nasal airflow errors were also reported in 19%-31% of Swedish 5-year-old children.[12,13] In 2001, Sell et al.[14] reported that 19% of UK 5-year-olds with unilateral cleft lip and palate had poor intelligibility and 18% had hypernasality. Since then, services in the UK have been centralized into high volume centres where care is delivered by specialist clinicians working in multidisciplinary teams, or at a local level by therapists often supported with specialist training and advice from the centre. Fifteen years on, CCUK reported on the outcomes of cleft care in this new model.[15–20] Although Sell et al.[18] reported improvements across several speech parameters, 17% of children still had poor intelligibility at 5 years of age within this centralized service.

There are a number of factors which have been identified in the literature that may have an impact upon speech outcomes in children born with cleft lip and palate. These include socio-economic status, gender, hearing, timing and method of primary palate repair, timing, method and amount of speech intervention, velopharyngeal insufficiency, fistulae and dental/occlusal anomalies.[11,21–28] While velopharyngeal insufficiency post primary palate repair is in itself a speech outcome, it can also have a significant impact upon articulation in this population. The importance of the above factors in relation to speech outcomes within a centralized (potentially more standardized) model of care is unclear.

Previous studies have often reported on numerous individual speech characteristics yet often these reports are more difficult for non-specialists to understand, in contrast to an overall global descriptor such as intelligibility. However the use of intelligibility alone as an outcome measure in cleft speech studies is controversial, with difficulties in its application, definition, measurement, the stimuli it is based on, and the nature of the listener.[29]A potential solution to this challenge is to report summary scores of overall articulation and structure alongside intelligibility. Factors contributing to a poor outcome for structure, that is velopharyngeal insufficiency and/or fistulae, may differ to those which contribute to a poor articulation outcome. Examining these two domains of speech outcome separately, that is structure and articulation, may enable more specific identification of the factors which affect them. In addition, some factors, such as velopharyngeal dysfunction or fistulae, may be causative in relation to, for example, the development and maintenance of speech articulation errors. Other factors may not cause the speech problem but may contribute to the persistence of errors, for example, limited access to speech intervention services, a lack of a stimulating environment, language delay or entrenched mislearning of speech sound patterns.[18,23,26]

Our aim was to describe centre-level variation in speech treatment and outcomes, and to explore factors associated with the poorest speech outcomes within a centralized service using data from CCUK.[17,18]

2 | METHODS

2.1 | Study sample

CCUK is a UK-wide cross-sectional study of 5-year-old children born between April 2005 and March 2007 with UCLP. A full description of recruitment procedures and eligibility criteria can be found elsewhere.[17,18] Briefly, of 359 eligible children, consent for participation was obtained from 268 (75%) children and parents. Ethical approval was obtained (REC reference number: 10/H0107/33, South West 5 REC). Eligible families were invited to attend a designated study clinic. Consent/assent was obtained from parents/children respectively to take part in this multi-outcome study on arrival at the clinic.

2.2 | Speech outcomes—general description

Two independent listeners analysed the speech recordings using the CAPS-A protocol.[18,30] The raw data from the CAPS-A assessment were used to derive three speech outcome variables: structure, articulation and intelligibility/distinctiveness. Scores were deduced from the ratings for each parameter and the colour system in CAPS-A. Each colour on the form was assigned a score, with dark green being 0, light green 1, yellow 2, and red 3. Colour coding indicated severity of outcome: green being a very good/ good outcome, red a poor outcome and amber in the middle.

2.3 | Speech—articulation outcome

The outcome related to articulation was derived from narrow phonetic transcription of speech recordings collected at assessment. The speech errors observed in the recordings were then coded into one of 11 cleft speech characteristics. Scoring of each cleft speech characteristic was based on the number of target consonants affected by the characteristic. Each summary category of the cleft speech characteristics (anterior, posterior, non-oral) was coded 0-3 reflecting the CAPS-A colour coding, as described in the Appendix. For each child the category (anterior, posterior and non-oral) with the highest score was used as their score for the articulation outcome variable.

2.4 | Speech—structural outcome

Outcomes related to structure (velopharyngeal insufficiency and/ or fistulae) included the following parameters: hypernasality, audible nasal emission, nasal turbulence and passive consonant errors. Each variable was coded 0-3 reflecting the CAPS-A colour coding, as described in the Appendix. For each child the parameter with the highest score was used as their score for the structural outcome variable.

2.5 | Speech—intelligibility/distinctiveness

The intelligibility/distinctiveness scale was scored on a 5-point ordinal scale (0 (dark green), 1 (light green), 2 (amber), 3 (light red), 4 (red). This was rated as a measure of the ability of an unfamiliar listener to understand speech, based on a short sample of conversational speech. This outcome was designed to assist the interpretation of results and was not to be used as a stand-alone speech result.[30]

2.6 | Factors associated with speech outcomes—general description

Variables that may be associated with speech outcomes were chosen based on the existing literature and availability in the data set. Variables were identified in the following categories: socio-demographic, parental happiness with their child's speech, hearing, speech intervention and surgical/dental. Each of these is described below and detailed in Table 1.

2.7 | Factors associated with speech—socio-demographic variables

Age and gender were recorded. Age at speech assessment was calculated using the child's date of birth. Information on the languages used in the home was obtained from parental questionnaire. The Index of Multiple Deprivation was used as a proxy of socio-economic position. This is a geographically based (postcode) relative measure of deprivation and consists of a weighted score covering up to seven domains (income, employment, education, skills and training, health and disability, crime, housing and living environment). Higher scores indicate higher deprivation. The score is used to rank neighbourhoods from most deprived to least deprived. We obtained deprivation ranks from England (http://geoconvert.mimas.ac.uk/help/imd-2007-manual.pdf), Scotland (http://www.gov.scot/Topics/Statistics/SIMD/SIMDPostcodeLookup/ScotlandPostcodeLookup) and Wales (https://statswales.gov.wales/Catalogue/Community-Safety-and-Social-Inclusion/Welsh-Index-of-Multiple-Deprivation/Archive/WIMD-2011). These neighbourhood ranks are subject to small changes over time and IMD scores go back to 2007, 2009, and 2011 for England, Scotland and Wales, respectively. We used ranks from these earliest records as they are closest to the year of birth and to the birth to 5-year exposure period of our cohort. The ranks are relative to other neighbourhoods within each country; they are therefore not comparable in an absolute sense between countries. To harmonize, we classified individuals in the lowest quartile within our cohort for each country as living in the most deprived areas.

TABLE 1 Description of sample[a]. All results are N and % unless stated

	N	n (%)
Age (years)	248	5.5 (5.4, 5.7)
Sex (female)	248	81 (32%)
Structure		
Green (0)		149 (61%)
Light Green (1)	243	47 (19%)
Amber (2)		2 (1%)
Red (3)		45 (19%)
Articulation		
Green (0)		154 (62%)
Light Green (1)	248	24 (10%)
Amber (2)		40 (16%)
Red (3)		30 (12%)
Intelligibility[b]		
0		134 (56%)
1		21 (9%)
2		42 (18%)
3		23 (10%)
4		18 (8%)
Deprivation score (percentile rank)[c]	223	40 (17, 66)
History of hearing loss/ OME (yes)	187	85 (45%)
Current hearing problems (best ear>20 dB)	194	51 (26%)
No SLT (yes)	238	49 (21%)
History of treatment for CSC (yes)	243	66 (27%)
Ongoing treatment for CSC (yes)	243	85 (35%)
Treatment required but not in therapy (yes)	243	67 (28%)
English is additional language (yes)	242	26 (11%)
Secondary speech surgery (yes)	244	62 (25%)
Fistula (yes)	244	42 (17%)
Muscle repair (yes)	176	53 (30%)
Fistula closure (yes)	228	21 (9%)
Unhappy with child's speech (0-6)	226	50 (22%)

OME, otitis media with effusion.
[a]Results are restricted to the 248 children with speech data.
[b]Intelligibility: rating on a 5-point ordinal scale of the ability of an unfamiliar listener to understand speech: 0 corresponds to no problems, 4 impossible to understand.
[c]Median and interquartile range: lower ranks indicate a relatively more deprived area.

2.8 | Factors associated with speech—parental satisfaction with speech

Data relating to parental happiness with their child's speech was assessed using an 11-point Likert-type scale where 0 equates to "very unhappy" and 10 equates to "very happy" with their child's speech. For this study, a score of 0-6 was used as a marker of parents being unhappy with their child's speech.[31]

2.9 | Factors associated with speech—hearing treatment and outcome measures

History of hearing and ENT problems were collected using a standardized proforma completed by an audiologist from parent interviews and medical records. Children were coded as having a history of otitis media with effusion and hearing loss if they had worn hearing aids, or had grommets or t-tubes inserted following palate repair up to the age of 5 years. Current hearing (at age 5) was assessed using pure tone audiometry by audiologists.[19] For this analysis children with hearing thresholds of >20 dB in their best ear were classified as having hearing loss. The hearing variables used in this paper are described in more detail in another paper in this supplement.[32]

2.10 | Factors associated with speech—speech and language therapy

The specialist centre-based speech and language therapists completed a questionnaire using information from parents, medical notes and local speech therapy services.[17] Information was gathered about current and past speech interventions. Variables used in the analysis were derived from these data and consisted of a categorical variable for whether intervention for cleft speech characteristics had been received in the past or not, and a variable relating to whether speech intervention was indicated but not being received at 5 years of age.

2.11 | Factors associated with speech—surgical and anatomical

Medical records were used to ascertain the surgical treatment of each child. Data on history of muscle repair at primary surgery, secondary speech surgery and fistula closure were gathered. The presence of a fistula at age 5 was based on a judgement made during the dental examination.

2.12 | Statistical analysis—agreement between scores

Cross-tabulations were used to understand the relationship between the three outcomes of structure, articulation and intelligibility/distinctiveness. Kappa agreement analysis was also performed after creating binary variables to classify children with the worst scores in each outcome (description below). Kappa values >0.75 were taken to reflect excellent agreement, those between 0.4 and 0.75 good agreement and those <0.4 moderate or poor agreement.[33]

2.13 | Statistical analysis—centre-level variation

Centre-level variation in treatment for speech and outcomes was examined using logistic multilevel models. Based on these models, we estimated the variance partition coefficient (VPC)—a measure of the proportion of total variation that can be attributed to centre, and used estimates from the model to predict the mean outcomes in each centre. Likelihood ratio tests were performed to assess whether any observed variation between centres could be attributed to chance. The treatment variables explored were: secondary speech surgery (yes/no); history of treatment for CSCs (yes/no) and if speech treatment was required but the child was not in therapy at the time of the recording (yes/no). The outcome variables assessed were articulation, structure and intelligibility, coded as binary variables as described below. All results were adjusted for differences in age and sex. A full description of these models is in Wills et al. (2017).[34]

2.14 | Statistical analysis—factors associated with speech outcomes

As our aim was to describe and investigate children with the poorest outcomes, we reclassified the ordinal articulation, structural and intelligibility/distinctiveness outcomes into binary variables where children who were scored as 3 for articulation and structure, and 3 or 4 for intelligibility/distinctiveness were classified as having a poor outcome for that variable.

For each factor, a logistic multilevel model was estimated with centre as a random effect and age and sex as fixed effects. All associations are expressed as odds ratios and reflect the odds of a poor outcome in the exposed vs unexposed categories, adjusting for age, sex and centre. We also re-estimated the between-centre variation (VPC) after including each predictor and calculated the change in VPC compared to the model not including the predictor (using the same sample). We used R (vers 3.3.2) and the lme4, blme and R2MLwiN packages package to estimate the VPCs. Stata (vers 14.2) was used for all other analyses.

3 | RESULTS

3.1 | Sample description

Speech and language recordings were available on 248 children. The recordings allowed the derivation of speech scores: 248 (93%) for the articulation outcome, 243 (91%) for the structural outcome and 238 (89%) for the intelligibility/distinctiveness outcome. Table 1 shows the results for each of these outcomes.

3.2 | Agreement between measures of speech outcomes

The associations between speech outcomes scores are shown in Table 2. Structural and articulation scores were associated with intelligibility/distinctiveness. For example, of the 42 children with a poor structural outcome and 26 children with a poor articulation outcome, only 9 (21%) and 4 (16%) respectively had no intelligibility issues (Table 2). The agreement between articulation score and intelligibility was good (κ=0.47), and the agreement between structural score and intelligibility was moderate

TABLE 2 Cross-tabulations of articulation, structure and intelligibility outcomes

		Articulation (%)					
		Green	Light Green	Amber	Red	Total	
Structure	Green	98 (65.8)	17 (11.4)	18 (12.1)	16 (10.7)	149 (100)	
	Light green	28 (59.6)	5 (10.6)	13 (27.7)	1 (2.1)	47 (100)	
	Amber	0 (0)	0 (0)	1 (50)	1 (50)	2 (100)	
	Red	25 (55.6)	2 (4.4)	8 (17.8)	10 (22.2)	45 (100)	
	Total	151 (62.1)	24 (9.9)	40 (16.5)	28 (11.5)	243 (100)	
		Intelligibility (%)					
		0	**1**	**2**	**3**	**4**	**Total**
Structure	Green	101 (69.1)	13 (8.9)	16 (11.0)	10 (6.8)	6 (4.1)	146 (100)
	Light green	27 (61.36)	13 (8.9)	10 (22.7)	3 (6.8)	0 (0)	44 (100)
	Amber	0 (0)	0 (0)	0 (0)	1 (50)	1 (50)	2 (100)
	Red	5 (11.9)	4 (9.5)	15 (35.7)	7 (16.7	11 (26.2)	42 (100)
	Total	133 (56.8)	21 (9.0)	41 (17.5)	21 (9.0)	18 (7.7)	234 (100)
Articulation	Green	108 (72)	7 (4.7)	23 (15.3)	7 (4.0)	4 (3.3)	150 (100)
	Light green	14 (60.9)	5 (21.7)	1 (4)	3 (13)	0 (0)	44 (100)
	Amber	10 (25.6)	7 (18)	14 (36)	7 (18)	1 (3)	39 (100)
	Red	2 (8)	2 (8)	4 (15)	6 (23)	12 (46.2)	26 (100)
	Total	134 (56.3)	21 (8.8)	42 (17.7)	23 (9.7)	18 (7.6)	238 (100)

(κ=0.33). Articulation and structural scores showed poor agreement (κ=0.15).

3.3 | Centre-level variation in speech outcomes and treatment

Results of the centre variation analysis are shown in Table 3 and Figures 1-6. There was strong evidence of centre-level variation in the intelligibility/distinctiveness outcome between treatment sites and a weak suggestion of some variation in the structural speech outcome. Approximately 13% and 5% of the total variation in intelligibility and structure, respectively, could be assigned to differences between centres. Table 3 also shows that there was evidence of variation in intervention for speech between centres, with approximately 15% and 9% of the total variation in secondary speech surgery and history of treatment for CSCs, respectively, being attributable to centre differences. Figures 1-6 show the predicted proportion in each centre and show that this variation was attributable to several centres.

3.4 | Factors associated with articulation outcome

Table 4 shows the factors associated with a poor articulation outcome. If a child had a fistula or a parent reported unhappiness with their child's speech, the odds of having a poor articulation outcome were approximately four- to fivefold higher. Children with a history of hearing problems were also more likely to have poor articulation

TABLE 3 Predicted mean with each outcome for the so-called average centre and the between-centre variability

Factor	n	Proportion (95% CI)	VPC[a]	*P*-value[b]
Treatment/ management				
Secondary speech surgery (yes)	260	0.25 (0.05, 0.35)	0.15	<.001
History of CSC treatment (Yes)	259	0.30 (0.10, 0.62)	0.09	.006
Treatment required but not in therapy (Yes)	259	0.38 (0.18, 0.62)	0.08	.10
Outcome				
Articulation (poor)	248	0.06 (0.02, 0.16)	0.05	.32
Structure (poor)	243	0.15 (0.05, 0.35)	0.05	.08
Intelligibility (poor)	238	0.12 (0.02, 0.48)	0.13	<.001

All results are adjusted for age and sex.
[a]VPC, variance partition coefficient.
[b]A test of the null hypothesis that there is no between-centre variation.

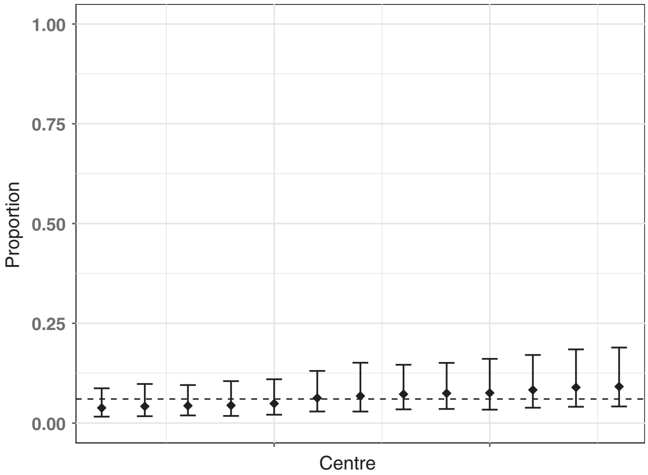

FIGURE 1 Predicted proportion of children with poor articulation in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

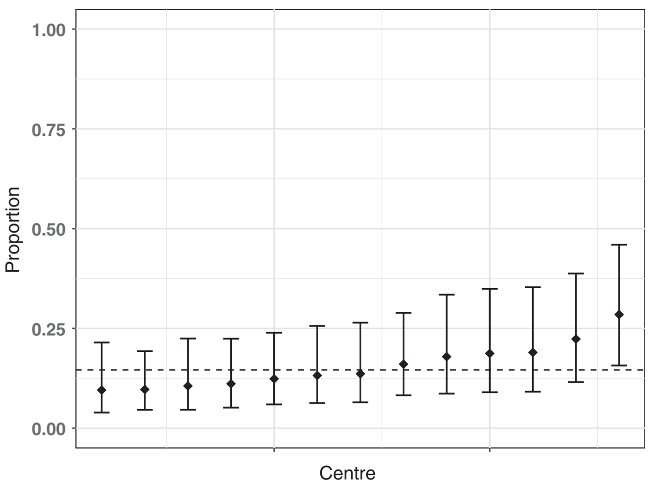

FIGURE 2 Predicted proportion of children with poor structure in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

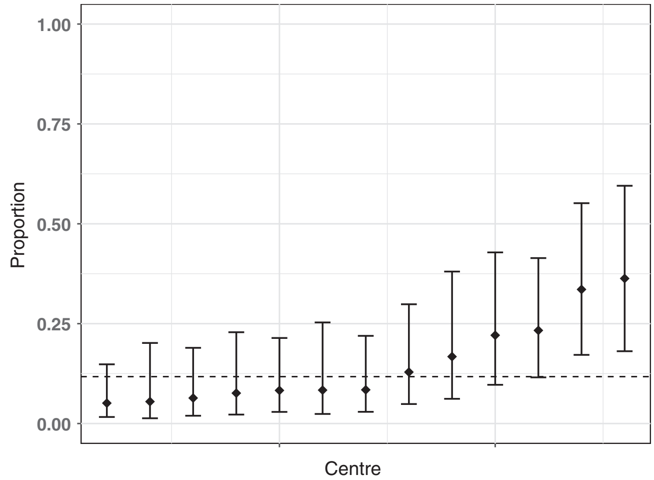

FIGURE 3 Predicted proportion of children with poor intelligibility in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

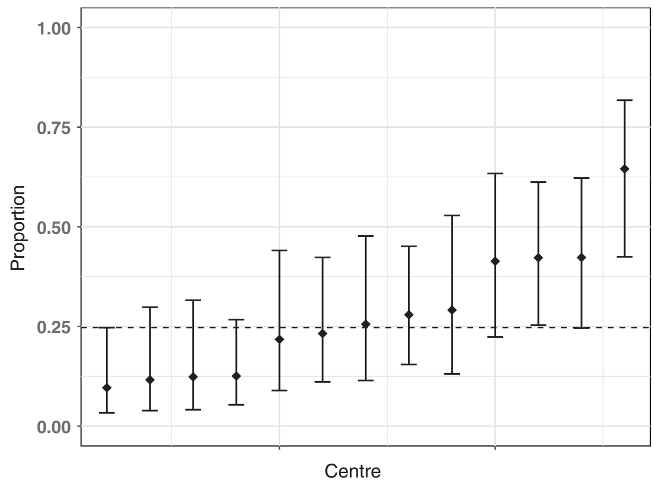

FIGURE 4 Predicted proportion of children who had received secondary speech surgery in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

and there was weak evidence that this outcome was more prevalent in boys. Children who had received treatment for cleft speech characteristics were almost three times less likely to have a poor articulation outcome. Variation in the structural outcome explained some of the variation in articulation between centres.

3.5 | Factors associated with structural outcome

Table 5 shows the factors associated with a poor structural outcome after adjusting for age, sex and centre. The odds of a poor structural outcome were twofold higher in parents who reported unhappiness with their child's speech. Children who had received secondary speech surgery had six times the odds of a poor structural outcome. Children with a history of treatment for cleft speech characteristics were approximately three times less likely to have a poor structural outcome.

3.6 | Factors associated with intelligibility/distinctiveness

Table 6 shows the factors associated with a poor intelligibility outcome. There was strong evidence of an association between parental unhappiness with speech and intelligibility/distinctiveness; children whose parents reported being unhappy with their speech had thirteen times the odds of a poor intelligibility outcome. Children with poor structural outcomes were also more likely to have poor intelligibility/distinctiveness. A history of therapy for CSCs again had a protective association with a poor intelligibility/distinctiveness outcome; only

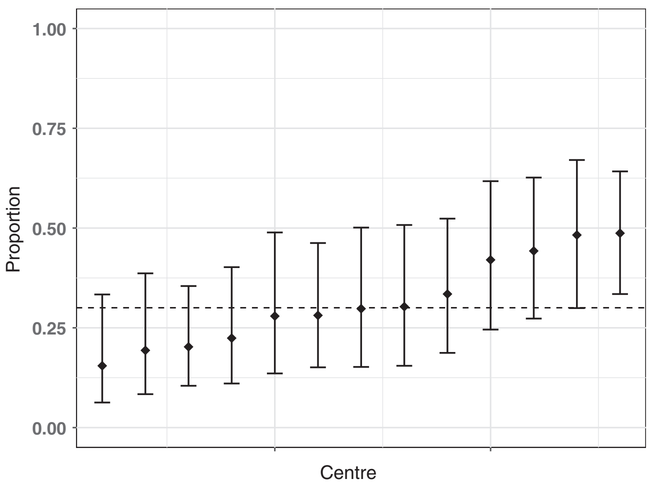

FIGURE 5 Predicted proportion of children who had received therapy for cleft speech characteristics in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

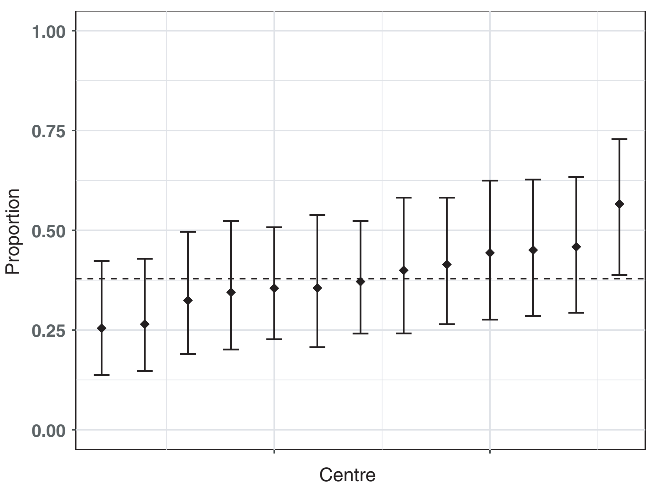

FIGURE 6 Predicted proportion of children who were identified as requiring treatment but were not in therapy in each centre. The bars are 95% confidence intervals and the dashed line is the predicted mean for the average centre. Adjusted for age and sex

one of the 62 children with a history of treatment had a poor intelligibility/distinctiveness outcome.

3.7 | Explaining centre variation

Muscle repair at primary surgery reduced the between-centre variation in articulation and intelligibility/distinctiveness by 3.5% and 8% in absolute terms, respectively. Likewise, secondary speech surgery explained approximately 4% of the between-centre variation in the structural outcome while therapy for cleft speech characteristics also explained a smaller amount of the variation in outcomes between centres (Tables 4-6). It was not possible to see how combining all of the treatment factors might reduce the total variation because the sample size reduced to 81 and estimation became problematic.

4 | DISCUSSION

There was variation in treatment and speech outcomes across centres within this centralized multidisciplinary service at age 5. Poor speech outcomes were associated with a fistula, previous secondary speech surgery and history of hearing loss, discussed in detail in the accompanying paper in this supplement.[32] Previous speech intervention was associated with a lower odds of a poor speech outcome. Parental concern about speech was also strongly associated with poor speech in the child.

4.1 | The value of different speech outcomes

Three measures of speech outcome were examined: articulation, that is presence of cleft speech characteristics in speech output; structural, that is features in speech which are indicative of velopharyngeal insufficiency or presence of a fistula; and intelligibility/distinctiveness, that is a measure of the ability of an unfamiliar listener to understand speech. There was only weak agreement between structure and articulation confirming they are measuring different aspects of speech but there was moderate-to-good agreement between these outcomes and intelligibility/distinctiveness suggesting that intelligibility/distinctiveness captures at least a part of these outcomes.

Intelligibility/distinctiveness was included as an outcome in CCUK to allow comparison with the previous pre-centralization survey.[18] However, because of several concerns about this measure, including the use of ordinal scales for its measurement and the lack of external validity, this scale has been removed from routine audit reporting in the UK.[34] Our findings that the structural and articulation scores were reflected onto the intelligibility/distinctiveness ratings and that associations were also seen with parental concern and history of therapy for cleft speech characteristics questions this decision and suggests that the CAPS-A intelligibility/distinctiveness measure may be valid and valuable.

To answer the research questions, it was necessary to reduce the speech data into two separate outcome variables of structure and articulation, rather than report on the individual subscores which are available from each of the measures. Others too have recognized the need to summarize speech data,[35–41] not least to simplify the reporting of speech outcomes for the multidisciplinary team and to reduce the number of variables in the statistical analysis. Similar to the structural score in our study, Lohmander et al[39]proposed the Velopharyngeal Composite Score (VPC-sum) and subsequently validated this. In two studies based on CAPS-A data, Pereira et al.[40] reported on a similar approach and found it to be a useful speech outcome, although recommended validation against another perceptual or instrumental measure of velopharyngeal function. In the second study based on CAPS-A data, Ahl et al.[36] reported on an overall VPD severity score based on the highest rating observed among resonance, nasal airflow, and passive cleft speech characteristics, similar to the approach adopted in our study. We selected this approach to be able to categorize the poor outcomes using the colour coding system.

TABLE 4 Associations between each factor and poor articulation. The change in between-centre variation (VPC) after including each treatment related factor is also shown[a]

Variable	N	OR	*P*	Change in VPC[a]
Male	248	2.56 (0.92, 7.15)	.073	
Most deprived (lowest quartile)	223	0.91 (0.32, 2.56)	.862	
History of hearing loss/ OME	187	2.68 (1.07, 6.70)	.035	+0.021
Current hearing problems (dB>20)	194	1.04 (0.40, 2.67)	.943	+0.001
History of treatment for CSC	243	0.29 (0.08, 1.02)	.053	+0.017
Treatment required but not in therapy	243	1.23 (0.50, 3.02)	.657	+0.008
English is additional language	242	0.33 (0.04, 2.66)	.296	
Secondary speech surgery	244	1.12 (0.44, 2.83)	.807	+0.007
Fistula at age 5	244	4.03 (1.67, 9.77)	.002	+0.02
Muscle repair	176	0.60 (0.17, 2.09)	.419	−0.035
Fistula closure	228	0.88 (0.18, 4.29)	.876	+0.002
Not happy with child's speech (0-6)	226	5.51 (2.26, 13.45)	<.001	
Structure (VPD score=3)	243	1.33 (0.95, 1.84)	.095	

All results are adjusted for age and sex
[a]The baseline VPC was 0.05. To make results comparable, the change in VPC was calculated by refitting the baseline model using the same sample.

TABLE 5 Associations between each factor and poor structure. The change in between-centre variation (VPC) after including each treatment related factor is also shown[a]

Variable	N	OR	*P*	Change in VPC
Male	243	1.39 (0.65, 2.99)	.394	
Most deprived (lowest quartile)	218	0.41 (0.15, 1.16)	.093	
History of hearing loss/ OME (yes)	183	1.12 (0.52, 2.41)	.767	+0.003
Current hearing problems (dB>20)	190	0.81 (0.34, 1.95)	.636	0
History of treatment for CSC	238	0.32 (0.12, 0.86)	.024	−0.024
Treatment required but not in therapy	238	0.74 (0.32, 1.67)	.466	−0.005
English is additional language	237	2.08 (0.70, 6.17)	.184	
Secondary speech surgery	239	6.19 (2.74, 13.98)	<.001	−0.041
Fistula at age 5	240	1.43 (0.60, 3.39)	.422	+0.014
Muscle repair	171	0.75 (0.29, 1.94)	.547	−0.005
Fistula closure	223	0.82 (0.22, 3.05)	.762	+0.001
Not happy with child's speech (0-6)	222	2.29 (1.08, 4.86)	.03	

All results are adjusted for age and sex
[a]The baseline VPC was 0.05. To make results comparable, the change in VPC was calculated by refitting the baseline model using the same sample

We also had concerns that the name of the outcome measure could be misleading. The use of VPC or VPD suggests that all the perceptual features are attributable to velopharyngeal insufficiency (VPI) but this may not be the case if there is a persistent fistula. Although nasal turbulence is only ever associated with VPI, other perceptual features, including audible nasal emission and hypernasality, do overlap for these two aetiologies, hence we preferred to use the term structural.

Unlike others, we have also developed the summary aspects for the articulation domain of CAPS-A, using the principle of taking the highest rating observed across the categories as the overall outcome. In addition, we did derive an overall summary speech score based on the combined structural and articulation scores which we anticipated might replace the intelligibility/distinctiveness measure. However, this outcome parameter did not show significant associations with the independent variables or account for any centre-level variation, in contrast to intelligibility/distinctiveness, and hence the detail of the overall summary score has been excluded from this paper.

4.2 | Centre-level variation in treatment and speech

The observed variation between centres may reflect differences in the need, differences in practice or differences in the resources available in each centre. For example, centre-level variation in secondary speech surgery is determined by multiple factors including parental choice, surgical protocol, differences in the team's threshold for treatment of velopharyngeal insufficiency,[42] and issues such as season and child's health and level of maturation. The differences in provision of therapy may reflect inequity in staffing levels of speech and language

TABLE 6 Associations between each factor and poor intelligibility. The estimates are adjusted for centre, age and sex. The change in between-centre variation (VPC) after including each treatment related factor is also shown[a]

Variable	N	OR	*P*	Change in VPC
Male	238	1.49 (0.62, 3.55)	.371	
Most deprived (lowest quartile)	214	1.06 (0.41, 2.69)	.91	
History of hearing loss/ OME (yes)	178	1.59 (0.68, 3.76)	.286	0.002
Current hearing problems (dB>20)	185	1.26 (0.48, 3.34)	.639	−0.002
History of treatment for CSC	233	0.05 (0.01, 0.37)	.004	−0.029
Treatment required but not in therapy	233	1.10 (0.47, 2.55)	.831	0.002
English is additional language	232	1.16 (0.28, 4.73)	.838	
Secondary speech surgery	234	1.67 (0.72, 3.85)	.232	0.029
Fistula at age 5	234	1.99 (0.80, 4.96)	.142	0.036
Muscle repair	168	0.45 (0.11, 1.81)	.261	−0.079
Fistula closure	219	0.43 (0.05, 3.68)	.444	0.017
Not happy with child's speech (0-6)	217	13.16 (4.93, 35.10)	<.001	
Structure (VPD score=3)	234	1.82 (1.34, 2.46)	<.001	

[a]The baseline VPC was 0.13. To make results comparable, the change in VPC was calculated by refitting the baseline model using the complete cases.

therapists and resources within the cleft centres and in local services, which is usually outside of the control of the centres.[43]

4.3 | Fistula and poor speech outcome

This study has found that a presence of a fistula at 5 years of age was associated with a poor articulation outcome. Henningsson and Isberg[44] and Lohmander et al.[45] attributed backing or retracted articulation to fistulae. This is where speakers change the place of articulation of consonants to that which is posterior to the fistula, to unconsciously avoid its consequences on speech. Even after fistula repair these erroneous habits persist, requiring speech intervention. Interestingly, the presence of a fistula was not associated with a poor structural outcome but this maybe because the structural outcome was not sensitive to the specific speech consequences of a fistula. Also in this study a straightforward measure of presence vs absence of fistula was used and a more detailed approach looking at size and location of fistulae might be more informative. A recent report however suggests there is poor inter-rater reliability when using more detailed fistula classifications.[46]

It is noteworthy too that backing or retracted articulation, that is posterior errors, is not restricted to occurring only in the presence of a fistula. Trost-Cardamone et al.[47] hypothesized that they may also be linked to a history of hearing loss, but it is not unusual for no physical explanation for this common error type in cleft palate speech.

4.4 | Secondary speech surgery and poor speech outcome

A history of secondary speech surgery was predictive of a poor structural outcome. This suggests that there were still features of frequent nasal emission and/or nasal turbulence, and/or passive speech characteristics, and/or moderate/severe hypernasality post-surgery. This is somewhat disappointing at first sight. However, it is well recognized that there is a subgroup of children with intrinsically poor levator muscles who have a poor prognosis for velopharyngeal closure following primary and secondary speech surgery, and revision secondary speech surgery may be required. Secondly, this finding may reflect the two-stage procedure which has been promulgated in the UK to avoid the consequences of an "unphysiological" pharyngoplasty.[48,49] The two-stage procedure may not have been completed by the age of 5 years. Thirdly, secondary speech surgery may have resulted in considerable improvement in reducing moderate or severe hypernasality, but there remains some residual nasal emission and/or nasal turbulence, about which families and clinicians are often unconcerned and often do not seek more intervention.[50] In this study there were nine children following secondary speech surgery who had oral tone but frequent nasal turbulence. Indeed, the CAPS-A nasal turbulence scale has drawn criticism for its sensitivity.[51,52] Yet another explanation may be that velopharyngeal closure has been successfully gained following secondary speech surgery but the structural outcome may be reflecting the consequences of a fistula. In summary, although these data suggest that secondary speech surgery, when considering group results, is not successful, to fully understand and interpret this finding, a separate analysis is planned of all of the children who received secondary surgery with an analysis of those variables which are associated with good and poor structural speech outcomes.

4.5 | History of speech intervention and poor speech outcome

Receiving therapy in the preschool years for cleft speech characteristics was associated with a lower odds of poor speech outcomes, although interestingly this contrasts with the findings of the Scandcleft study in which large amounts of speech intervention were associated with a poor speech outcome.[53,54]

While intervention for cleft speech characteristics logically improves the articulation and intelligibility/distinctiveness outcomes, it is less intuitive how therapy also provided some beneficial effect to the structural outcomes. However, it is known that where there is velopharyngeal mislearning, for example active nasal fricatives, glottal and pharyngeal articulation, these error types are associated with an open velopharyngeal sphincter with speech consequences suggestive of a structural impairment.[55,56] Speech intervention addressing the mislearning may reduce the impact of the velopharyngeal insufficiency, and the resulting structural scores. In summary, our study is highly supportive of speech intervention for cleft speech characteristics in the preschool years. Nevertheless, even within the centralized model of care in the UK, there are inequities and shortages in speech and language therapy provision.[43] This inequity of provision has been raised at a national level and is the subject of an ongoing study. Alternative models of speech therapy delivery may be indicated, to achieve better access to speech intervention. For example, as in speech disorders of non-cleft origin,[55,56] it may be that parents can be trained to support the development of speech in their children.[57]

4.6 | Parental concern and poor speech outcome

Our findings suggest that parents are highly attuned to their children's speech and that parental concern could be a useful proxy for speech status where more formal assessments of speech are unavailable. These findings are consistent with reports from other studies that have drawn attention to the importance of parental concerns[58,59] and shown how parental concern is associated with poorer outcomes.[8]

4.7 | Other factors associated with poor speech

Gender is commonly associated with speech outcomes in the non-cleft population[60] with boys being at greater risk but gender differences have not always been considered in cleft speech studies. Our finding that poor articulation is likely to be more prevalent in boys is consistent with the Scandcleft study,[53] who also reported that boys had lower scores than girls on their articulation measure. In contrast, measures of deprivation were not strong predictors of speech outcomes in this data set, a similar finding to the study by Choa et al.[61]

4.8 | Strengths and limitations and recommendations for future research

This study was a large study (for a study of children with cleft lip and palate), nationwide with a good response rate and based on a validated measure of key outcomes measured with enough precision to demonstrate improvements over time. However, this study does have a number of limitations. Missing data was an issue when analysing several factors across different domains. A deeper exploration of some of the factors included here is also required to provide further insight into the associations observed, for example, details of secondary speech surgery and its impact on speech, and whether a more detailed system for classifying fistulae size and location is feasible. Other variables should be investigated relating to primary surgery, including individual surgeon and their experience, surgical techniques and timing of repair. Future analysis on this and other data sets should explore centre variation in the different domains of speech outcomes, and their associations with different patterns of service provision. Future studies need to be larger and longitudinal to investigate the development of speech and the factors associated with improved outcome. Like Lohamnder et al.[54] and Willadsen et al.[53], this study also indicates a need to understand much more fully speech intervention, including the optimum timing, method and amount of SLT intervention in order to know better how to make best use of resources and achieve best possible outcomes.

5 | CONCLUSIONS

Poor speech outcomes in children born with UCLP vary across centres in the UK, with the greatest variation in intelligibility/distinctiveness. Centres vary in treatment of velopharyngeal insufficiency and provision of speech intervention. Poor speech outcomes at age 5 years are associated with a fistula, previous secondary speech surgery and history of hearing loss. However, previous speech intervention mitigated against a poor speech outcome. Parental concern about speech is also strongly associated with a poor outcome and may be a useful indicator of speech when more formal measures are unavailable. These findings support the importance of early identification and management of fistulae, effective management of velopharyngeal insufficiency and hearing impairment, and most importantly access to speech intervention in the preschool years, in the context of the growing body of evidence that persistent speech disorders at 5 years of age have significant long-term consequences.

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APPENDIX

Deriving Structural, Articulation Scores and an Overall Summary Score based on the CAPS-A Tool.

The highest score for each of the two outcomes of structure and articulation are deduced from the ratings and the colour system, where dark green=0, light green=1, amber=2 and red=3. The structural summary score is derived from the scales of hypernasality, audible nasal emission, nasal turbulence and the passive category. The articulation summary score is derived from the anterior, posterior and non-oral categories. The highest value from any of the variables within the category of structure or articulation was used as the child's outcome score for that domain.

Structural Score

Variable label	Variable value	Outcome
Hypernasality	3	Red (3,4)
	2	Amber (2)
	1	Light Green (1)
	0	Green (0)
Audible nasal emission	2	Red (3)
	1	Light Green (1)
	0	Green (0)
Nasal turbulence	2	Red (3)
	1	Light green (1)
	0	Green (0)
Weak and or nasalized (passive CSC)	3	Red (3)
	1,2	Amber (2)
	0	Green (0)
Nasal realizations (passive CSC)	3	Red (3)
	1,2	Amber (2)
	0	Green (0)
Gliding (passive CSC)	3	Red (3)
	1,2	Amber (2)
	0	Green (0)

Articulation Summary Score

Variable label	Variable value	Outcome
Lateral/ Lateralization (Anterior CSC)	3+ consonants affected	Amber (2)
	1, 2 consonants affected	Light Green (1)
	0 consonants affected	Green (0)
Palatal/ Palatalization (Anterior CSC)	3+ consonants affected	Amber (2)
	1, 2 consonants affected	Light Green (1)
	0 consonants affected	Green (0)
Double articulation (Posterior CSC)	3+ consonants affected	Amber (2)
	1, 2 consonants affected	Light Green (1)
	0 consonants affected	Green (0)
Backed to velar (Posterior CSC)	3+ consonants affected	Red (3)
	1,2 consonants affected	Amber (2)
	0 consonants affected	Green (0)
Pharyngeal (Non-oral CSC)	3+ consonants affected	Red (3)
	1,2 consonants affected	Amber (2)
	0 consonants affected	Green (0)
Glottal (Non-oral CSC)	3+ consonants affected	Red (3)
	1,2 consonants affected	Amber (2)
	0 consonants affected	Green (0)
Active nasal fricative (Non-oral CSC)	3+ consonants affected	Red (3)
	1,2 consonants affected	Amber (2)
	0 consonants affected	Green (0)
Double articulation (Non-oral CSC)	3+ consonants affected	Red (3)
	1,2 consonants affected	Amber (2)
	0 consonants affected	Green (0)

Within the categories of posterior and non-oral, the following should also be interpreted as “red” outcomes.

POSTERIOR

Where the two cleft speech characteristics (ie double articulation and backed to velar) are **both** coded as amber, this was scored as a red outcome for articulation.

NON-ORAL

Where there are two or more cleft speech characteristics (ie pharyngeal, glottal, active nasal fricatives, double articulation) coded as amber, this was scored as a red outcome for articulation.