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Centre-level variation in dental treatment and oral health and individual- and area-level predictors of oral health in 5-year-old children with non-syndromic unilateral cleft lip and palate: the Cleft Care UK study. Part 3

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Structured Abstract

Objectives: To explore centre-level variation in fluoride treatment and oral health outcomes and to examine the association of individual- and area-level risk factors with dental decay in Cleft Care UK (CCUK).

Setting: Two hundred and sixty-eight 5-year-old British children with non-syndromic unilateral cleft lip and palate (UCLP).

Materials and Methods: Data on caries and developmental defects of enamel (DDE) were collected. The child’s history of fluoride ingestion and postcode was used to assess exposure to fluoridated water. Centre-level variation in fluoride exposure and caries was examined using hierarchical regression. Poisson regression was used to estimate the association between individual- and area-level fluoride exposures and outcome.

Results: Children had high levels of caries, rampant caries and DDE. There was no evidence of variation between centres in the number of children with caries or rampant decay. There was evidence of variation in prescription of fluoride tablets and varnish and the type of toothpaste used. Area level of deprivation was associated with a higher risk of dental caries—risk ratio (RR) in the lowest quartile versus the rest was 1.43 (95% CI 1.13 to 1.81). Use of fluoride tablets and varnish was associated with higher risk of caries—RR 1.73 (95% CI 1.29 to 2.32) and RR 1.33 (95% CI 1.04 to 1.70), respectively, adjusted for age, sex and deprivation.

Conclusion: The association with use of fluoride tablets and varnish probably reflects reverse causality but indicates the need for early preventative interventions in children with UCLP.

KEYWORDS
caries, centralization, cleft lip, cleft palate, fluoride, oral health, variation

1 | INTRODUCTION

Many studies have reported that children with cleft lip and palate have poorer oral health outcomes. Our previous research examined the impact of centralization on oral health in 5-year-old children with unilateral cleft lip and palate (UCLP) in the Cleft Care UK study. We found that there had been no improvement in oral health following centralization of services with a mean number of decayed missing and filled primary teeth (DMFT) increasing by 0.60 (95% CI 0.00 to 1.21). This is an open access article under the terms of the Creative Commons Attribution License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited.
teeth (dmft) of 2.3. By comparison, the most recently published UK Child dental Health (CDH) survey reported that the average dmft in 5-year-olds was 0.9. No studies have investigated whether oral health treatment and outcome varies between centres in a centralized model of care.

There are a number individual risk factors in both tooth structure and environment that influence oral health. These include socio-demographic factors (such as deprivation and ethnicity), behavioural factors (such as diet and dental hygiene), treatment factors (such as applied topical treatments) and structural defects (such as developmental anomalies and quality of the enamel). In the general population, area-level risk factors for poorer oral health outcomes include relative deprivation and low water fluoride levels. Few studies have examined the association between individual- and area-level factors and oral health in children with cleft lip and palate.

In this study, analysis of Cleft Care UK (CCUK) is extended to explore centre-level variation in fluoride treatment and oral health outcomes, to describe patterns of decay and to examine the association of area- and individual-level risk factors with dental decay.

## METHODS

### 2.1 | Study sample

Data from CCKU were used. This 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. 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).

### 2.2 | Oral health measures

The collection or oral health data has been described in detail previously. Briefly, information on each child's oral health was recorded using a standardized proforma. Data on caries were collected by consultants in paediatric dentistry who had completed a training and calibration programme based on the British Association for the Study of Community Dentistry (BASCD) criteria for caries assessment and a modified developmental defects in enamel (DDE) index for assessing dental anomalies of the upper incisors. Some children in particular centres were found to have had their dental assessment performed by uncalibrated assessors (n=69). All the available clinical records and photographs were reviewed by a calibrated paediatric dentist (JS) and any discrepancies recoded. DDE was unrecordable for a further six children.

### 2.3 | Oral health outcomes

Using the decayed, missing, filled teeth (dmft) format, levels of caries and the treatment received for caries were recorded for each one of the primary teeth. Children were defined as having severe or extensive dental decay (rampant caries) if they had: five or more teeth with obvious decay experience (dmft of 5+); three or more teeth with decay into dentine (new or recurrent); any very severely decayed teeth, deemed "unrestorable"; evidence of sepsis or any teeth extracted due to decay. DDE data were recorded for the primary incisor teeth.

### 2.4 | Individual fluoride exposure measures

Parents were asked about their child’s history of fluoride ingestion. Information on fluoride tablets or drops (yes/no), whether fluoride varnish had been applied by a dentist (yes/no) and the type of toothpaste the child was using at the time of the study was gathered (coded as children’s: 1000 ppm, adult: 1450 ppm or other).

### 2.5 | Water fluoridation measures

Information on water fluoridation was gathered by linking residential postcodes to reports on water fluoride content from their local water authority. Mean fluoride levels (mg/L) could be ascribed for 263/268 (98%) children. This was converted to a categorical variable for analyses using the following threshold levels: low <0.3, medium 0.3<0.7, high >0.7 mg/L. These thresholds have been used widely in the dental literature.

### 2.6 | Socio-demographic variables

Age and sex were recorded. Age at dental assessment was calculated using the child’s date of birth. The Index of Multiple Deprivation (IMD) was used as a proxy of socio-economic position. This is a geographically based (postcode) relative measure of deprivation and comprises 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://geocovans.mimas.ac.uk/help/imd-2007-manual.pdf), Scotland (http://www.gov.scot/Topics/Statistics/SIMD/SIMDPopcodeLookup/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.

### 2.7 | Statistical analysis—centre-level variation

Centre-level variation in fluoride treatment, fluoride content of toothpaste, dmft and rampant decay was examined using hierarchical regression models. Variation in DDE score between centres was not analysed because this outcome is not affected by dental treatment,
rather it is a reflection of cleft severity. 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. All results are adjusted for differences in age and sex. Full details of the method for examining centre-level variation is described in Wills et al. (within this supplement). As noted above, some children in particular centres had their dental assessment performed by uncalibrated assessors, and all children from one particular centre were assessed by an uncalibrated observer. This may bias assessments of centre variation as the uncalibrated observers tended to either consistently over- or underestimate the prevalence of disease (as verified by the revalidation carried out by author JS). As a sensitivity analysis, the centre variation models were refitted after excluding those individuals whose measurements were not performed by a validated assessor.

2.8 | Statistical analysis—associations with dmft, rampant decay and DDE

Two sets of Poisson regression models were used to estimate the association between each of the individual- and area-level fluoride exposures and each outcome. Poisson rather than logistic regression was used so that risk ratios could be calculated—odds ratios are difficult to interpret when the outcome is common such as in this study (Table 1). The first set of models adjusted for age (years) and sex (a minimally adjusted model), and the second was additionally adjusted for deprivation (percentile rank). For all Poisson models, we used robust standard errors to control for mild violations of the model assumptions. Stata v14.2 was used for all analyses.

2.9 | Statistical analysis—sensitivity analysis

The robustness of findings to the quality of the outcome assessment was investigated by repeating analyses after restricting to the observations from the calibrated observers that had also been revalidated by the consultant paediatric dentist (JS) (n=189/264, 72%). Potential bias from missing data was also explored by refitting the minimally adjusted models on the complete cases. For both these sensitivity analyses, findings were broadly similar (see Tables S1 to S3 in supplementary material) and so the results presented use all available data regardless of revalidation.

3 | RESULTS

3.1 | Sample description

Table 1 shows the characteristics of children in CCUK. Approximately two-thirds of the children were boys. Children were, on average, living in slightly more deprived areas of the United Kingdom (median percentile rank: 41).

<table>
<thead>
<tr>
<th>TABLE 1 Characteristics of CCUK sample a</th>
</tr>
</thead>
<tbody>
<tr>
<td>N</td>
</tr>
<tr>
<td>---</td>
</tr>
<tr>
<td>Male</td>
</tr>
<tr>
<td>Age (median, IQR)</td>
</tr>
<tr>
<td>Deprivation (median, IQR) b</td>
</tr>
<tr>
<td>Caries present</td>
</tr>
<tr>
<td>dmft (mean)</td>
</tr>
<tr>
<td>dmft if dmft&gt;0 (mean)</td>
</tr>
<tr>
<td>Untreated Caries</td>
</tr>
</tbody>
</table>

**DDE**<sup>b</sup> (n of teeth)

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>264</td>
<td>125 (47.4%)</td>
</tr>
<tr>
<td>1</td>
<td></td>
<td>69 (26.1%)</td>
</tr>
<tr>
<td>2</td>
<td></td>
<td>52 (19.7%)</td>
</tr>
<tr>
<td>3</td>
<td></td>
<td>14 (5.3%)</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>4 (1.5%)</td>
</tr>
<tr>
<td>DDE≥1</td>
<td>257</td>
<td>139 (52.7%)</td>
</tr>
<tr>
<td>Rampant decay</td>
<td>260</td>
<td>75 (28.9%)</td>
</tr>
<tr>
<td>Fluoride tablets prescribed</td>
<td>245</td>
<td>25 (10.2%)</td>
</tr>
<tr>
<td>Fluoride varnish applied</td>
<td>254</td>
<td>60 (23.6%)</td>
</tr>
<tr>
<td>Toothpaste fluoride content</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;1000 ppm</td>
<td>232</td>
<td>104 (44.8%)</td>
</tr>
<tr>
<td>1000 ppm +</td>
<td>128</td>
<td>55.2%</td>
</tr>
<tr>
<td>Mean concentration of fluoride in water</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;0.3 mg/L</td>
<td>259</td>
<td>216 (83.4%)</td>
</tr>
<tr>
<td>0.3 to 0.7 mg/L</td>
<td>14</td>
<td>5.4%</td>
</tr>
<tr>
<td>&gt;0.7 mg/L</td>
<td>29</td>
<td>11.2%</td>
</tr>
</tbody>
</table>

aData are presented for the 264/268 that had dental health assessment.

bDDE, Developmental defects in enamel.

3.2 | The prevalence of oral disease and individual- and area-level risk factors

The mean dmft was 2.3, but in children with decay, the mean dmft was 4.2. Seventy-five children (29%) had rampant caries, and 138 children (54%) had at least one incisor with a DDE. Eighteen per cent of the children had at least one filling, and 10% had at least one tooth extracted. Ten per cent and 23% of children had been prescribed fluoride tablets and treated with fluoride varnish, respectively, and 30% (73/242) had received at least one of these treatments. Only 29 (11%) children were registered in a district with a high-fluoride water content, the majority (84%) were living in districts with low levels of fluoride (<0.3 mg/L).

3.3 | Centre-level variation in fluoride exposure and oral health outcomes

Table 2 shows the results of the between-centre variability analyses. There was no evidence of variation between centres in the number of children with caries or rampant decay. However, there was substantial variation in the use of fluoride tablets and varnish between centres.
Approximately 57% of the variation in fluoride tablets could be apportioned to centres. Figure 1 shows the estimated proportions in each centre; only four sites had prescribed tablets, and one site in particular contributed to over half of the total prescriptions in the United Kingdom in our sample. Approximately 9% of the variation in fluoride varnish was attributable to centre differences. Almost all centres had prescribed varnish to at least one child in our sample, but three centres had higher rates (Figure 2). Prescription of fluoride tablets was strongly associated with the presence of a paediatric dentist at the centre \((P = .009)\)—no tablets were prescribed at centres where there was no paediatric dentist. Children were also more likely to receive fluoride varnish if the cleft team included a paediatric dentist, although the evidence was equivocal \((27\% \text{ v } 16\%, P = .12)\).

Fluoride content of toothpaste used by children also varied by centre. Approximately 36% of the total variation lay between centres (Table 2), and there was evidence that this variation went in both directions with some centres having lower and some centres having a higher prevalence of children using toothpaste with higher fluoride content (Figure 3).

<table>
<thead>
<tr>
<th>Outcome</th>
<th>n</th>
<th>Proportion (95% CI)</th>
<th>VPC</th>
<th>(P)-value*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Caries (yes)</td>
<td>264</td>
<td>0.57 (0.42, 0.70)</td>
<td>0.02</td>
<td>.34</td>
</tr>
<tr>
<td>Rampant decay (Yes)</td>
<td>260</td>
<td>0.24 (0.15, 0.37)</td>
<td>0.02</td>
<td>.9</td>
</tr>
<tr>
<td>Fluoride tablets (Yes)</td>
<td>245</td>
<td>0.0005 (0.00, 0.97)</td>
<td>0.57</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Fluoride varnish (Yes)</td>
<td>254</td>
<td>0.25 (0.07, 0.58)</td>
<td>0.09</td>
<td>.02</td>
</tr>
<tr>
<td>Toothpaste(^a) (1000 ppm+)</td>
<td>232</td>
<td>0.68 (0.13, 0.97)</td>
<td>0.26</td>
<td>&lt;.001</td>
</tr>
</tbody>
</table>

\(^a\)Fluoride content of toothpaste.

\(^*\)The \(P\)-value is a test of the null hypothesis that there is no between-centre variation. All results are adjusted for age and sex.

VPC, Variance partition coefficient.
3.4 Individual and area-based measures and dental caries

There was no centre variation in dental caries at age 5 (Figure 4). The association between fluoride exposure and area level of deprivation with dental caries and severe or extensive dental decay is shown in Table 3. Children who had been prescribed fluoride tablets or varnish had a higher risk of caries and severe or extensive dental decay. There was strong evidence that children living in deprived areas were more likely to have caries and weak evidence of a similar association with severe or extensive dental decay. The increase in risk was 46% (95% CI: 13 to 89%) and 59% (95% CI: 0 to 52%) for caries and severe or extensive dental decay, respectively.

3.5 Individual and area-based measures and developmental enamel defects

Table 3 shows the association between DDE and fluoride exposure and area level of deprivation. There was no association with individual or area levels of deprivation. There was weak evidence that children using higher fluoride-level toothpaste had a lower risk of enamel defects.

4 DISCUSSION

There was a high prevalence of caries, severe or extensive dental decay and enamel defects in this population of children with cleft lip and palate. The proportion of children receiving preventive treatments like varnish was low. There was no centre-level variation in oral health outcomes, but there was centre-level variation in fluoride treatment and in the fluoride content of toothpaste that a child used. Living in a fluoridated area was not associated with dental caries, but area-level deprivation was associated with a higher risk of dental caries as was the use of fluoride tablets and varnish.

4.1 Prevalence of caries and rampant caries

Our data suggest that the prevalence of oral health outcomes was higher than in the general population. The prevalence of dmft >0, and severe or extensive dental decay was 54% and 29%, respectively. It is of relevance and a useful comparison that the most recently published UK CDH survey reported that the prevalence of dmft >0 and rampant caries in 5-year-old children was 31% and 13%, respectively. Our findings of higher risk of dental caries is consistent with other studies. Various reasons such as slower oral clearance of food and the greater incidence of hypomineralized teeth have been suggested as the cause of this increased caries susceptibility.

4.2 Prevalence of developmental enamel defects

The reported prevalence of DDE in primary teeth varies from 4% to 40% in different populations. In our population, this was higher (at 54%) and this may represent an underestimate as children with missing teeth could not be scored. Other studies have reported a higher prevalence of DDE in children with clefts. The aetiology of DDE is not entirely clear, but it has been associated with poor maternal health during pregnancy, pre-term birth and hospitalization in the first year of life; the majority of these children would have been admitted to hospital in their first year of life for cleft repair operations. There is scant evidence as to why children born with a cleft have a high prevalence of DDE but a case control study looking at permanent teeth showed colour changes (which would include DDE, mild fluorosis, early dental caries) of enamel three times more likely in these children. There is likely upset to enamel formation in the developmental stages, and this manifests in these colour changes. It has been speculated that the disturbance to enamel may represent an incomplete manifestation of the clefting process. Nevertheless with more than 90 known influences on enamel formation, it is difficult to establish the absolute causes of alterations seen in the enamel.

4.3 Centre-level variation in treatment and outcome

There was cross-centre variation in prescription of fluoride tablets and fluoride varnish applications; our results suggest this may reflect the differences in staffing of units where having a paediatric dentist increases the likelihood of these treatments being prescribed. There was no variation in oral health outcome within this centralized multidisciplinary service. In some centres, examiners were not calibrated but further validation work using photographs that has been validated in previous studies and sensitivity analyses suggest that this would not have disguised large differences.
4.4 | Deprivation and dental caries

Children from deprived backgrounds had more chance of having caries and rampant caries. Deprivation has been associated with a higher consumption of non-milk extrinsic sugars and with lesser use of fluoride toothpastes, both factors associated with a greater caries risk.26,27 This is consistent with findings from other studies in the general population28,29 and suggests that deprivation is an important risk factor in children with cleft lip and palate.30

4.5 | Water fluoridation and dental caries

The effectiveness of water fluoridation has been shown in epidemiological studies and cessation studies where community water fluoridation was withdrawn.31,32 Area-level measures were available in this study but assumed that the child had lived at the address long term and made no allowance for consumption of bottled water. This imprecision in exposure measurement together with the low power (10% to detect a difference in those exposed to
fluoridation) may explain why no protective association was observed.

4.6 | Fluoride toothpaste, fluoride treatment and dental caries

A Cochrane review that pooled 75 studies confirmed that toothpaste with concentrations of 1000 ppm prevents dental caries in children. As the majority of parents reported that their children used 1000 ppm or 1450 ppm toothpaste and may have reported this inaccurately, it is not surprising that we were unable to observe a protective association. Fluoride varnish application has been shown to reduce caries in randomized trials and population programmes in Scotland and Wales. Similarly, long-term regular provision of fluoride tablets or drops reduces risk of caries. Use of fluoride varnish and tablets was low in this study with only 24% and 10% of families, respectively, reporting their child as having received this treatment. Furthermore, use of tablets and varnish was associated with a higher (rather than lower) level of caries that likely represents reverse causality and suggests that fluoride varnishing was used as a control measure once disease was identified rather than as a purely preventative measure.

4.7 | Strengths and limitations

This study was a large (for a study of children with cleft lip and palate), nationwide with a good response rate and a series of validated measures of key outcomes measured with enough precision to demonstrate improvements over time. However, there are a number of limitations. First, this study has limited power to detect modest centre-level variation in treatment and outcome. Second, the fluoride history was recorded from the accompanying adult. They were asked about fluoride sources potentially available to their child including toothpaste, fluoride supplements in the form of drops or tablets, professionally applied fluoride varnish and water fluoride. These questions were asked in the same format as the previous cross-sectional study to allow comparability but rely on the knowledge, understanding and memory of the adult. They were also only required to give a yes/no response in some circumstances, therefore giving no information on whether exposure was short or long term. Most toothpastes available at the time of data collection contained 1000 ppm or 1450 ppm sodium fluoride. In general, toothpaste designed for children 3 and under has 1000 ppm and for older children and adults 1450 ppm, but not all parents will be aware of the fluoride level in the toothpaste being used. Third, not all dental examiners were calibrated although validation using photographs and sensitivity analyses suggested this will not have distorted the findings though it may have reduced power. Fourth, we had too few children from different ethnic groups to explore this issue and we had not data on other important exposures such as intake of dietary sugars.

4.8 | Research implications

Future studies need to be larger and longitudinal with repeated measures of oral health and treatment to better describe the determinants and sequelae of poor oral health in children with cleft lip and palate, for example identifying hypomineralized teeth at an early age and following the fate of each individual tooth to determine the role of hypomineralization in caries development in cleft children.

We have shown that children with cleft lip and palate in the United Kingdom are now treated by a centralized multidisciplinary service. However at that time these children were born, there was very limited access to paediatric dentists within most regional units. Although other aspects of the delivery of cleft care were able to develop very quickly after centralization, the resource to develop paediatric dental services was not immediately apparent and varied between centres. This is difficult to measure precisely and may be an important factor as to why centralization did not show improved oral health at age 5 in our study. Indeed, the prevalence of dental caries and developmental enamel defects is high and the use of preventative measures of proven effectiveness is low.

Further work is required to develop the optimal early intervention package to prevent dental caries in children with cleft lip and palate and study the outcome of implementation of this package. There are few examples or models to follow, but as dental caries is a wholly preventable disease, it is important that a strategy is developed as part of a key outcome of the CCUK study.

5 | Conclusions

Children with cleft lip and palate in the United Kingdom are now treated by a centralized multidisciplinary service, but at the time these children were born, there was very limited access to paediatric dentists within most regional units; this may be an important factor explaining why centralization did not show improved oral health at age 5 in our study. Indeed, the prevalence of dental caries and developmental enamel defects is high and the use of preventative measures of proven effectiveness is low. The only centre-level variations identified were in the use of fluoride tablets and varnish, measures prescribed by dentists. The association of these with high caries rates suggests that they were prescribed when caries was already identified. There was no variation in treatment or outcome suggesting that the service provided as these children were developing was uniformly failing to provide preventative programs and that national improvements in service provision are required.

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