Identifying the effect of cleft type, deprivation and ethnicity on speech and dental outcomes in UK cleft patients: A multi-centred study

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Summary  Background: Outcome measures are increasingly important in the modern National Health Service. In the care of children born with cleft lip and/or palate there are many different outcomes to consider but only a few reliable, validated outcome measures exist.

The dmft (decayed, missing and filled teeth) index and cleft speech characteristics (CSCs) are used regularly by cleft teams throughout the UK to assess outcomes in children with cleft lip and/or palate. We hypothesized that these two outcome measures might be significantly influenced by the demographics of the populations studied independent of the care provided.

Methods: A retrospective review of all patients aged between five and six referred to three regional cleft centres during a twelve month period were included in the study. Speech and dental outcomes were compared with patient ethnicity, cleft type and level of deprivation as determined by the Carstairs score.

Results: The data of 287 patients were used. Speech was significantly affected by cleft type (p < 0.03), whereas dentition was significantly affected by ethnicity (p = 0.002) and deprivation (p = 0.012).

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Conclusions: This study demonstrates that the demographics of cleft populations can significantly affect the measures of outcome used to assess the quality of care provided by cleft teams. It has also demonstrated that these demographics are not evenly distributed across the country and that some cleft teams will have a more ‘at risk’ population than others.

Level of evidence: Risk, level II.

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Introduction

Outcome measures are increasingly important in the modern National Health Service (NHS). The 2010 government white paper entitled ‘Liberating the NHS — Equity and Excellence’ states: "Providers will be paid according to their performance. Payment should reflect outcomes, not just activity, and provide an incentive for better quality". However, outcomes may be influenced not only by the quality of care provided but also by the demographics within the populations affected by disease.

In the care of children born with cleft lip and/or palate there are many different outcomes to consider and only a few reliable, validated outcome measures exist. Decayed, missing and filled deciduous teeth can be assessed using the British Association for the Study of Community Dentistry (BASCD) criteria at five years of age — dmft index. The dmft index is used to assess dental caries based on three values: the number of deciduous teeth with carious lesions, the number of extracted teeth, and the number of teeth with fillings. The Cleft Audit Protocol for Speech — augmented (CAPS-A) is an assessment tool for use in inter-centre audit studies of cleft speech. It includes ratings of both nasal airflow errors and cleft speech characteristics (CSCs). CAPS-A scores for CSCs can be summarised as shown in Table 1.

Dmft and cleft speech characteristics are used regularly by cleft teams throughout the UK to assess outcomes in children with cleft lip and/or palate. We hypothesized that these two outcome measures might be significantly influenced by the demographics of the populations studied independent of the care provided.

<table>
<thead>
<tr>
<th>Table 1</th>
<th>CAPS-A scoring for cleft speech characteristics.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft speech characteristic score</td>
<td>Features</td>
</tr>
<tr>
<td>0</td>
<td>No CSCs</td>
</tr>
<tr>
<td>1</td>
<td>Very minor differences, anterior CSCs only.</td>
</tr>
<tr>
<td>2</td>
<td>Non-oral CSCs, backing to velar</td>
</tr>
<tr>
<td>3</td>
<td>Passive CSCs/non-oral CSCs</td>
</tr>
</tbody>
</table>

Methods

Three UK regional cleft centres, West Midlands, South West/South Wales, and Spires collect and compare outcome data annually for inter centre audit. The outcomes compared at these annual meetings include dmft and CSCs in children aged five years. This study examined the records collected on the cohort of children born in 2004 with cleft lip and/or palate. Records were collected between the patients’ fifth and sixth birthdays. All children referred to each centre and born in 2004 for whom there were records available were included in the study. Demographic data collected included gender, ethnicity and the postcode of the patient.

Ethnicity was determined from the self-reported information collected by the hospital administration systems. Ethnic minority groups were collapsed to provide four groups with larger numbers of patients — Caucasian, Mixed, Asian and Other.

The postcode available on the hospital administration system for each patient at the time of the study was used to calculate a level of deprivation. There are a number of methods to measure deprivation; the chosen method for this study was the Carstairs index, developed by Carstairs and Morris in the 1980s. It is based on four census indicators: low social class, lack of car ownership, overcrowding and male unemployment. Geographical areas are divided according to their postcode based on these variables. High scores indicate a high level of deprivation, with the whole range of scores being divided into quintiles.

The cleft type determined at the time of the first operation was recorded for each patient. The type of cleft was subdivided into five groups: unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), cleft palate only (CP), cleft lip and alveolus (CLA) and cleft lip alone (CL).

Dental caries for all children with a cleft lip and/or palate as described by dmft was scored by appropriately BASCD calibrated consultants in paediatric dentistry in each cleft team. The only children excluded from dmft scoring were those who could not cooperate with the examination and those who were not brought to the clinic between their fifth and sixth birthdays.

Standardised speech recordings were made for all children with a cleft palate who were brought to the clinic between their fifth and sixth birthdays. Recordings were then assessed by CAPS-A trained listeners using the CAPS-A system. Consensus listening was used for all children scored
and independent listening was used for ten percent of the cases assessed. Children were excluded from speech assessment if their primary surgery had taken place in a different centre, if they had a diagnosed syndrome, if they were unable to complete the full data collection and if they were not seen between their fifth and sixth birthdays.

Patients with incomplete data in one category e.g. ethnicity, were not excluded from the whole study if their remaining data could contribute to another category.

Statistical methods

Results were analysed with the Pearson chi-squared test and Fisher’s exact tests and non-parametric tests including the Kruskal–Wallis for comparing the medians of the groups.

Results

The data of 287 patients were used in the study. Demographic data can be seen in Table 2.

Gender

There were no significant differences in gender distribution between centres and gender did not affect outcomes.

Ethnicity

In total, ethnicity data was available for 274 of the 287 patients. The range of ethnicities between the three centres is shown in Figure 1. Data from the South West and South Wales centre are represented separately throughout the graphs. Caucasian ethnicity is the predominant ethnicity in all centres and all patients recruited from the South Wales centre were Caucasian. The greatest ethnic diversity was seen in the West Midlands where 71% of patients in the study were Caucasian. Data for patients who were of mixed race were not included in the statistical analyses as the small number in this group (eight) would not have produced meaningful conclusions. However, analysis of the remaining groups shows there is a statistically significant difference ($p < 0.001$) between the centres in the ethnic diversity of their caseloads.

Table 2  Demographic data.

<table>
<thead>
<tr>
<th></th>
<th>South West</th>
<th>Spires</th>
<th>South Wales</th>
<th>West Midlands</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of patients</td>
<td>38</td>
<td>101</td>
<td>29</td>
<td>119</td>
<td>287</td>
</tr>
<tr>
<td>Male/Female</td>
<td>17</td>
<td>16</td>
<td>20</td>
<td>7</td>
<td>161</td>
</tr>
<tr>
<td>Gender</td>
<td>5</td>
<td>0</td>
<td>2</td>
<td>1</td>
<td>8</td>
</tr>
<tr>
<td>unrecorded</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Asian</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>19</td>
<td>21</td>
</tr>
<tr>
<td>Caucasian</td>
<td>32</td>
<td>81</td>
<td>27</td>
<td>84</td>
<td>224</td>
</tr>
<tr>
<td>Mixed</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>6</td>
<td>8</td>
</tr>
<tr>
<td>Other</td>
<td>0</td>
<td>11</td>
<td>0</td>
<td>10</td>
<td>21</td>
</tr>
<tr>
<td>Ethnicity</td>
<td>5</td>
<td>6</td>
<td>2</td>
<td>0</td>
<td>13</td>
</tr>
<tr>
<td>unrecorded</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
caries, whereas scores of one or greater indicate the child has active dental caries or has previously had primary teeth extracted or filled. In this study the dmft scores were used in a binary fashion when making comparisons with the variables under review — i.e. patients were considered to have caries experience with any score above zero. Figure 4 shows a trend of increasing incidence of caries experience across the different cleft types. A statistically significant relationship between dental caries and cleft type is not found if the cleft types are considered unrelated ($p < 0.096$). However, if it is assumed that the order of cleft types shown in Figure 4 represents an increasing order of severity of cleft, then the test for linear trend in percentage of non-zero dmft with increasing severity is significant ($p = 0.01$).

The level of deprivation of the patient was significantly associated with dmft scores ($p = 0.012$). As deprivation levels increased, the percentage of patients with a dmft score of one or more increased markedly as seen in Figure 5.

A statistically significant relationship was also demonstrated between ethnicity and dental outcomes ($p = 0.002$), with Asian patients having significantly worse outcomes than patients in either the Caucasian or ‘Other’ groups (Figure 6).
Cleft speech characteristics (CAPS-A)

Speech outcomes are only available for children who were born with a cleft palate. The likelihood of cleft specific characteristics in the speech of children with a cleft palate at age five years was found to be significantly related to cleft type \( p < 0.03 \) Figure 7. A more severe cleft was associated with increased cleft specific characteristics. Increased nasality in speech outcomes showed a similar trend but this trend was not significant.

However, cleft speech characteristics were not found to be significantly related to either deprivation scores \( p = 0.49 \) or ethnicity \( p = 0.15 \). The distribution of patients in each of the CSC categories for each deprivation quintile is too similar to show a significant association between CSCs and level of deprivation. In contrast, the speech data for different ethnicities results in small numbers in some subgroups making moderate variation between subgroups unlikely to be statistically significant.

In summary, the significant relationships between the variables of cleft type, deprivation and ethnicity and the outcomes of dental caries (dmft) and speech (CSCs) can be seen in Table 3 below.

Discussion

There is a wealth of literature that focuses on aetiological factors for cleft lip and palate (CLP).\(^6\) In addition, much is written on the outcomes of CLP patients relating specifically to the surgical intervention that the patient underwent.\(^7\) \(^9\) This study takes a different perspective, and investigates the effects of ethnicity, deprivation and cleft type on aspects of speech and dental outcomes in CLP patients.

This study demonstrated a similar distribution of cleft types across the different providers of cleft care involved despite differences in levels of deprivation and ethnicity between the centres. The distribution of cleft types in this study is similar to the national picture of distribution of cleft types for the UK as reported in the CRANE annual report 2010.\(^10\) Previously published studies from Scotland and Wales using the Carstairs and Townsend deprivation scales respectively have demonstrated an association between levels of deprivation and the prevalence of orofacial clefts.\(^11\)\(^12\) They did not however show a differential effect of deprivation on cleft type which is consistent with this study.

Cleft type however has been reported to be an important variable in the outcomes recorded for patients with cleft lip and/or palate. Previous publications have looked at aspects of the cleft to see how these affect speech outcomes. Hardin Jones\(^13\) and Persson et al.\(^14\) showed that children with clefts affecting the hard and soft palate required more speech therapy, had more articulation errors and a higher incidence of velopharyngeal incompetence than those with a cleft of the soft palate only. Sullivan et al.\(^15\) and Mahoney et al.\(^16\) showed that increasing width of the cleft resulted in increased rates of VPI. In contrast, Khosla et al.\(^17\) looked at the effect of cleft type on the rates of velopharyngeal incompetence in children treated with a Furlow palatoplasty and found no differences. In the study reported here, the type of cleft of each child was described and the frequency of cleft specific characteristics (CAPS-A) was significantly associated with cleft type. CSCs were more frequently seen in children with cleft types\(^14\) which are usually wider clefts and would usually be considered more severe such as a bilateral cleft lip and palate.

In this study dental caries experience was also related to cleft type, and increasing caries experience was related to increasing severity of the cleft. This is consistent with previous studies in the literature which have also looked at the effect of cleft type on dental caries.\(^18\)\(^19\)\(^20\) The results of these studies have shown differences in dmft scores between the different cleft types. Patients with combined cleft lip and palate have been shown to have significantly worse dental caries in the primary dentition than children with a cleft lip only.

The study reported here showed that the distribution of cleft types did not differ between units, but levels of deprivation and diversity of ethnicity did differ significantly between the units. In this study deprivation level and ethnicity are shown to be important variables when considering dental caries experience, but did not have an effect on speech outcomes. We did not identify other studies in the literature specifically investigating the effect of either ethnicity or deprivation on cleft outcomes.

However, deprivation has been shown to be strongly linked with dental caries in the non-cleft population.\(^21\) A survey of over 1800 parents in Leeds revealed significantly more caries with higher rates of deprivation using the Townsend deprivation score.\(^22\) Higher deprivation was linked with less frequent dental visits (including parental dental attendance) and use of community dental services. We did not identify studies specifically investigating the effect of deprivation on cleft dental caries experience.

Table 3  Demonstrating the effects of cleft type, deprivation and ethnicity on dmft and CSCs.

<table>
<thead>
<tr>
<th></th>
<th>dmft</th>
<th>CSCs</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft type</td>
<td>( p = 0.096 )</td>
<td>( p &lt; 0.03 )</td>
</tr>
<tr>
<td>Deprivation</td>
<td>( p = 0.012 )</td>
<td>( p = 0.49 )</td>
</tr>
<tr>
<td>Ethnicity</td>
<td>( p = 0.002 )</td>
<td>( p = 0.15 )</td>
</tr>
</tbody>
</table>
However it is reasonable to assume that the deprivation factors which lead to dental caries in the general population are also applicable to cleft patients from deprived backgrounds.

Studies of dental caries in non-cleft children in the literature have also shown that caries experience varies between different ethnic groups. Significantly higher rates of caries have been found in children from South Asia when compared with Caucasian children. This effect remains when fluoridation and deprivation using the Townsend index are controlled for. The current study also found a significant effect of ethnicity on dmft scores of children with a cleft lip and/or palate, with children of Asian origin having significantly higher dmft scores than children categorised as either Caucasian or ‘other’ origin.

This study has demonstrated that the demographics of cleft populations can significantly affect the measures of outcome used to assess the quality of care provided by cleft teams. The study has also demonstrated that these demographics are not evenly distributed across the country and that some cleft teams will have a more ‘at risk’ population than others. If cleft centres are to be compared using outcome measures it is important that such demographics are identified so that appropriate risk adjustment can be carried out when comparisons are made across centres. Identification of such risk demographics could also be used to identify more needy populations and influence resource allocation to help address such differences across the country.

**Limitations of the study**

Carstairs deprivation scores were calculated based upon the postcode of the patient at the time of record collection. Postcodes are subject to change over the five year period of the children growing from birth, diagnosis, and primary surgery to record collection. Also, ethnicity was taken as that recorded at the time of registration by administrative staff, and hence may not have been fully accurate.

Data collection and analysis of records collected was carried out by different people in the different cleft centres. However, the outcomes chosen for this study, dmft and CSCs, are both recorded by trained and calibrated practitioners. The possibility of bias in the recording of outcomes is therefore limited.

This study uses the cleft type as described at the time of initial surgery. Cleft type is not exactly equivalent to cleft severity. Other studies have used different methods of classifying the cleft of each child such as the Veau classification, and some studies have measured cleft width. This makes comparison across studies difficult.

There are other variables that will affect outcomes that have not been investigated in this study.

**Ethical approval**

Ethical approval was not deemed to be necessary for this study due to the retrospective nature of data analysis undertaken.

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**Conflict of interest statement**

None declared.

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**References**


