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Link to published version (if available): 10.1111/ocr.12355

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Title

Range and timing of surgery and surgical sequences used in primary repair of complete unilateral cleft lip and palate: The Cleft Care UK Study

Abstract

**Objects** - To describe the range of surgery used to repair the lip and palate in the UK with specific interest in the sequence/timing used in complete unilateral cleft lip and palate (cUCLP).

**Setting and Sample Population** - The Cleft Care UK study, a cross-sectional study of 268 5-year-olds, born from 2005-07, with complete unilateral cleft lip and palate.

**Materials & Methods** - Information on surgery was extracted from medical notes by surgeons during research clinics and transcribed onto a standardised questionnaire.

**Results** - Surgical data were available for 251 (94%) children from all cleft centres in the UK (n = 18). Over a two-year period, 32 surgeons used 10 different surgical sequences in primary repair of the cleft lip and palate. The most frequently used sequence was repair of cleft lip and anterior hard palate followed by repair of posterior hard palate and soft palate (70%). Four surgical sequences were used only once. Most surgeons had a preferred sequence, but 38% (11/29) used more than one sequence during the study period. The timing of repair of the lip, the hard palate and the soft palate varied with surgical sequence, and also between surgeons, even adjusting for the different sequences used.

**Conclusion** - Despite centralisation of cleft services in the UK, there remains considerable variation in both the sequence and timing of surgical repair of cleft lip and palate in infancy. Further work is required to understand whether these factors are associated with differences in outcome.
Keywords: cleft, surgery, cleft lip and palate, surgical workload

Introduction

The Cleft Care UK (CCUK) study is a United Kingdom (UK)-wide cross-sectional study of 5-year-old children born with complete Unilateral Cleft Lip and Palate (cUCLP)\(^1\). Recently reported findings from CCUK showed that outcomes in speech and dento-alveolar and facial form following treatment of cUCLP in the UK have improved since services were reorganised to a more centralised model of care\(^2^,\)\(^3\). This reorganisation of cleft services in the UK achieved a more multidisciplinary and audited service with designated cleft surgeons performing at least \(~40\) primary cleft repair operations (including all cleft types) per year\(^4\). However, in the UK centralisation process, neither surgical techniques nor the timing of primary surgery were standardised.

Previous studies of children with cleft lip and palate have found differences in outcomes between centres using different protocols of care\(^5^,\)\(^6^,\)\(^7\). The impact of protocols of care in such studies is unclear\(^5^,\)\(^7\) however. Some studies suggest that different techniques and protocols of care can result in similar outcomes\(^5^,\)\(^7\), and that differences in outcome may lie more in differences in surgical volume or skill\(^7^,\)\(^8^,\)\(^9^,\)\(^10\).

In previous publications a protocol usually refers to a surgeon’s or centre’s preferred order for surgical repair of lip, hard and soft palate, and alveolus, and the timing of that surgery (age of the child at surgery). It may also include the presence/absence and timing of other treatments such as pre-surgical orthodontics, and the technique used for each operation (eg Millard lip repair, Von Langenbeck palate repair).
The Eurocleft study[1] provides surgical information on all children as well as a protocol for each centre. The protocol for Centre A is lip repair at 3-5 months, soft palate repair at 9-18 months, and hard palate repair at 8-11 years. However, the surgical details indicate that 8% of children had lip and soft palate repair at the same time, and 23% a one stage palate repair, therefore not following the designated protocol for Centre A. Similarly, variation in sequence and timing from the designated protocol can be seen for Centres C and D. Such variations from the stated protocol limit the conclusions that can be drawn from comparisons based on the protocols used.

This report uses data from the CCUK study[1]. Our aims were to (a) describe the full range of different surgical sequences and timings (age at surgery) used to repair cleft lip and palate in infancy across all centres in the UK,(b) describe how consistently surgeons used surgical sequences and timings in their practice, and (c) estimate the extent of differences in timing of surgery. We also looked at surgical workload.

Methods
Study design and sample
CCUK is a UK wide cross-sectional study of 5-year-old children born between April 2005 and March 2007 with cUCLP. Data were collected from all cleft centres in the UK (n=18) at designated audit clinics. Out of 359 eligible children, consent for participation was obtained from 268 (75%) parents or guardians. The data used for this analysis came from the surgical questionnaire, and information was available from 251 (94%) of the children recruited to the study. The surgical questionnaire was completed during the special audit clinics by the surgeon using information from the medical notes. Additional information about the design and conduct of the CCUK study has been previously described[1]. Ethical approval was
Surgical variables

Surgical sequence

Seven exclusive options for specifying surgical sequence were provided in the surgical questionnaire along with an “other” free text category (see online supplementary material). For 11 cases the response given in the “other” category matched the options in the form and therefore these cases were assigned to the pre-defined option. Based on the responses in the “other” category additional categories were also formed where more than two children had a similar sequence. Two more sequences were created containing a total of 10 children.

Timing of operations

Information on the date of operation was combined with the date of birth to calculate the age of the child at each surgery. The questionnaire contained two sections that recorded this information. The first was the date given for each stage of the surgical sequence used, and the second was the date of operation for each of lip repair, lip adhesion, hard palate and soft palate surgery recorded. Where the dates from the two sections did not agree, we used the earliest date to calculate age at surgery (6, 7, and 8 cases for lip surgery, hard palate and soft palate repair respectively).

Hard and soft palate surgery were considered separately in this study. Hard palate surgery was considered to have taken place where the description of surgery included vomer flap, anterior hard palate, or hard palate, and the earliest surgical date mentioned was used for the analysis.
There were only eight cases of lip adhesion reported. This information was therefore pooled with the lip repair surgery variables to create a new variable which we refer to as age at primary lip surgery, where the earliest of the two types of surgery was analysed.

Age values that were zero or negative were removed. Based on these criteria, seven observations for age at lip repair, one value for hard palate and one value for soft palate were removed. An age was also considered implausible and removed if it was not possible based on the described sequence. Using these criteria, a child with Sequence B (1.lip 2.hard palate+soft palate) and a child with Sequence A (1.lip and anterior hard palate 2. posterior hard palate and soft palate) were removed as the dates of the lip, soft and hard palate surgeries were not concordant with the performed sequences.

**Surgeon and grade**

The name and grade (consultant or trainee) of the first named surgeon performing the operation was recorded. For the analysis the surgeons’ names were anonymised by assigning numerical codes.

**Other variables**

We also present summary statistics for gender, dento-alveolar relationship and deprivation score. A full description of these variables has been reported elsewhere\(^1\).

**Analysis**

Medians and inter-quartile ranges, and frequencies and percentages were used to describe the characteristics of the sample. Tests of difference between those in the analysis sample versus those eligible but missing data on the surgical variables were performed using Chi-square for
categorical variables and t-tests for continuous variables. Descriptive statistics of surgical sequence, and timings of surgery, and surgical volume were produced. We also stratified by grade of surgeon (trainee/consultant) since surgical sequence is decided by the lead consultant. Since there were only three trainees and they performed only 10 (<2%) of the operations, we restricted the rest of the analyses to consultant surgeons.

Linear random effects models were used to estimate the between-surgeon variation in timing of surgery. A random intercept was included for surgeon and the variance partition coefficient (VPC) was estimated. The VPC takes values between zero and one, and in our example captures the proportion of variation that can be attributed to surgeon. Two models were estimated for each of age at primary lip surgery, primary hard palate surgery and primary soft palate repair. The first model was unadjusted, and the second model was adjusted for surgical sequence. Plots of the predicted mean age at surgery (months) for each surgeon in the unadjusted and adjusted models were produced. Sensitivity analyses were carried out to check the influence of outliers. All statistical analyses were conducted using Stata v15.1.

Results

Description of sample

Out of the 251 (94%) children that had some information from the surgical questionnaire, one individual was excluded from the analysis because they did not have a complete cleft, and eight (3%) children were excluded because their questionnaire contained more than one surgical sequence. Three individuals had no information on the variables that are part of these analyses, leaving 238/268 (89.2%) children that were included in at least one of the analyses in this report. Table 1 shows descriptive statistics of those included in at least one
analysis and those eligible but excluded. There was no evidence that children eligible but not part of the analyses due to missing data were different to those included with respect to gender, neighbourhood deprivation, or dento-alveolar relationship (although the number for the latter was too small for a meaningful comparison). There was evidence of a difference in age at survey between those analysed versus those with some missing data although this difference was very small (mean difference: 2.2 months; 95\% CI: 0.4 to 4.0 months).

**Surgical volume by grade and surgeon**

Surgical volume is represented as the number of procedures that each surgeon performed within the two-year period for the 238 children included in the study. A procedure was defined as repair of lip, hard palate or soft palate. Two or more procedures may have been undertaken during the same operation. In total there were 32 different first named surgeons, only three (9.4\%) of whom were trainees in cleft surgery. Consultants performed approximately 98\% of the operations.

The number of procedures performed by each consultant surgeon by procedure type is shown in Figure 1 (the statistics to produce this figure are in Table S1 of the online supplementary material). Over the two-year period the median number of procedures performed by consultant surgeons was 23. Approximately one-third of consultant surgeons (9/28) performed more than 25 procedures, while approximately two-thirds of consultant surgeons (20/28) performed more than 15 procedures. Fifteen procedures over a two-year period is equivalent to caring for two to three children with the cleft type, cUCLP, per year. Each consultant surgeon did similar numbers of lip, hard palate and soft palate procedures.

**Surgical Sequence**
Information for surgical sequence was available from 31/32 surgeons. The frequency of each surgical sequence and median age of the child is shown in Table 2. The most common sequence was a lip and hard palate operation followed by posterior hard and soft palate repair. None of the children received sequences G (1. soft palate 2. lip and hard palate) and H (1. lip adhesion 2. lip repair 3. palate repair).

The timing of surgery was linked to the sequence. For example, compared to Sequences A and B, the hard palate operation occurred almost 3 months later in children that had sequence C, and the oldest ages at surgery were observed for Sequence E which comprised three operations (1. lip repair 2. soft palate 3. hard palate).

Figure 2 shows the number and variation of sequences performed by each consultant surgeon (the statistics to produce this figure are in Table S2 of the online supplementary material). Trainees have been excluded form Figure 2 as it is assumed that the surgical sequence was determined by the consultant. Consultant surgeons generally preferred a particular sequence, but many (38%) used more than one approach over the two-year period.

**Timing of surgery and variation in timing between-surgeons**

Table 3 and Figure 3 show the results from the analysis to estimate the between-surgeon variation in timing for all procedures. There was evidence of between-surgeon variation in timing for all procedures. Most variation was initially found for hard palate repair where 68% of the variation in timing was attributable to surgeon. However, a large part of this was explained by surgical sequence and one outlying surgeon. After adjusting for sequence the VPC reduced to 31% (95% CI: 14 to 56%), and this further reduced to 17% after removing the extreme outlying surgeon (surgeon no 12 in Fig 3B) who had a predicted mean timing of...
hard palate surgery much later (31 months) than the others (Table 3). The between-surgeon variation in timing of lip surgery and soft palate repair changed little after adjustment for surgical sequence - the amount of variation that could be attributed to differences between surgeons was 17% for lip surgery and 26% for soft palate surgery.

Without adjusting for surgical sequence, the range of predicted mean timings of surgery for each surgeon was 3.4 to 4.6 months for primary lip repair, 4.3 to 11 months for hard palate repair (excluding the outlier), and 6.1 to 11 months for soft palate repair. After adjusting for sequence, the range of predicted mean timings of surgery for each surgeon was broadly similar - 3.4 to 4.6 months for primary lip repair, 6.1 to 10 months for hard palate repair (excluding the outlier), and 7 to 11 months for soft palate repair. Adjusting for sequence affected the predicted mean timing of surgery for a small number of surgeons (Figures 3ABCD) because certain surgeons favoured particular sequences (Figure 2) and sequence was linked to timing of surgery (Table 2). However, the effect of adjusting for sequence on each surgeon’s predicted mean timing of surgery was generally small. The predicted mean timing for each surgeon was within the overall average except for 11 surgeons (33% of surgeons that operated on 110 (48%) of children) where there was evidence that surgery tended to occur earlier or later than by the average surgeon.

**Discussion**

**Summary of findings**

To the best of our knowledge, this is the first UK nationwide study to describe the full range of surgical sequences and range of timing of surgery used in the treatment of children with cUCLP. Ten different surgical sequences were used by 29 consultant surgeons. Some consultant surgeons always used the same sequence, but more than a third varied their
approach over the two year time period of the study.

There was variation in timing of surgery, particularly of the hard palate repair, that depended on the sequence used, and variation in timing of surgery between surgeons even when using the same surgical sequence.

**Surgical workload**

Over the period of the study the majority of children born with a cUCLP in the UK had their surgery carried out by a consultant surgeon. Consultant surgeons in this study had a wide range of workload with the busiest surgeon performing over fifty procedures on children with a cUCLP over this two-year period, and the least busy only two. In 2005-2007 two thirds of consultant surgeons appeared to be caring for at least two to three children with cUCLP per year or more, and the highest volume operators five to eight children with this cleft type each year. The rate of recruitment in this study varied across centres, with not all eligible children being included in the study. The figures in this study will therefore underestimate the number of children treated per surgeon. The Clinical Standards Advisory Group (CSAG) cleft study\(^\text{10}\) (prior to centralisation of cleft services in the UK) estimated that the majority of cleft surgeons at that time were looking after only 1-2 children with cUCLP per year, and the so called high volume operators were treating approximately six children with this cleft type per year. The results of this report are consistent with other studies finding that UK cleft surgeons have increased their cleft workload since centralisation of services\(^4,12\).

Although increasing the number of cases per surgeon per year was one of the recommendations of the CSAG study, there is currently little and conflicting evidence for improvement in cleft outcomes with increased volume of surgery\(^7,9,13,14,15\). In a specialty
where the prevalence of the condition is low and outcomes are often described over many years, understanding the effect of surgeon volume and experience on outcome is difficult. However, in other specialities a higher volume of operations and increased specialisation amongst surgeons have been associated with better outcomes.\textsuperscript{16, 17}

**Variation in surgical sequences and timing**

Ten different surgical sequences were identified for the primary repair of the cleft in children with cUCLP in the UK over a two-year period. None of these sequences included repair of the alveolus before the age of six years. Four of the sequences were only used once. Seventy percent of the children had closure of the cleft using the same sequence, namely repair of the cleft lip and the anterior hard palate first, followed by repair of the rest of the palate at a second stage.

The different sequences affected the timing of surgery to the lip, hard and soft palate, but surgeons also showed variation in timing of surgery when using the same sequence. Therefore variation in timing of surgery depended both on the sequence of surgery and the surgeon. However, there was a degree of consistency across the UK with all the children having their lip repaired by the age of six months, and all but three having their hard and soft palate repaired by the age of 13 months. The three exceptions were all operated on by the same surgeon who was an outlier in terms of timing of palate repair. Hard palate repair in these three children was delayed until after the age of two years.

Timing of surgery has been found to affect outcomes\textsuperscript{18, 19} but the ‘best’ timing of surgery is not known. This study shows that surgeons in the UK differ in the ages at which they perform primary cleft surgery. The differences in number of months are relatively small,
however, and it is not clear whether such differences affect outcomes.

**Strengths and limitations**

Not every eligible child in the UK during the study period was recruited to the study and recruitment from each centre varied. However, data are from a UK-wide cross-sectional study of 5-year-olds over a two-year period from 2010 to 2012, every cleft centre in the UK participated, and it recruited a large proportion of children of a similar age undergoing treatment at the time of the study. Findings are thus likely to be generalisable at least to national practice between 2005 to 2009.

Although this study examines surgical practice ten years ago, this data is new and provides a baseline against which future studies can be compared. The results of this study also suggest that investigation of the effect of early treatment on outcomes requires analysis of surgical information for each child, rather than analysis of protocols of care, as it is possible that not all children will be treated according to a stated protocol.

There are limitations to the study. The study calculated age at surgery from the date of birth. No allowance for gestational age was made, whereas the surgeon may take into account gestational age when planning a date for surgery. This is likely to be most significant in affecting the date of the first operation, cleft lip repair, with a delay in the first operation to accommodate for any prematurity of the infant. However, there is no reason to suppose that prematurity occurs more for one centre or surgeon than another, so the effect on surgeon variability would be expected to be small.

Information on the surgeons’ reasons for their choice of surgical sequence was not collected.
Neither were reasons for departing from their usual surgical sequence. Our report cannot therefore provide an understanding of the reasons for surgeons preferring one surgical sequence to another, or for changing sequence. All children in the study were non-syndromic, but data was not collected for instance on infant health, weight gain or family preferences on surgical timing, all of which may influence timing of surgery. We do not know if surgeons who always kept to the same sequence were simply inflexible, or felt it was better to always use the same sequence. Surgeons who used more than one sequence, on the other hand, may have felt that they used the sequence best suited to each particular child, or may have altered their sequence when they thought a case particularly easy or difficult, or may have simply liked to vary their approach.

While the information on surgical timing and sequence we report is comprehensive, important information was still omitted. For example, details of how the lip, hard palate and soft palate surgery was performed were not asked for. In addition, the dates of each surgical procedure were abstracted from hospital records by one surgeon during the audit clinic. There was no procedure to check this data in the study and data therefore could have been reported incorrectly. Since only a small number of children excluded from the analysis due to inconsistent data it seems unlikely that this is a major source of bias.

Future work

This study is the first of its kind to show the full range of surgical sequences and timings across all the UK cleft centres. Further work is being undertaken to determine whether the differences in surgical sequence, timing of repair of each part of the cleft, and the volume of surgery undertaken by a surgeon influence the outcome of the surgery.
Conclusions

Cleft surgeons in the UK use a range of different surgical sequences to repair the cleft lip and palate, and do not all consistently use the same sequence. There is variation in timing of closure of the lip, hard and soft palate cleft which is affected not only by the surgical sequence used but also by the individual surgeon. Investigation of the effects of surgery on outcomes in cleft care needs to take this into account.

References:


11. Semb G, Brattstrom V, Molsted K, Prahl-Andersen B, Shaw W. The Eurocleft Study: Intercenter Study of Treatment Outcome in Patients With Complete Cleft Lip and


Tables:

Table 1. Description of sample in those children used in at least one of the analysis presented and in those eligible but missing information on surgical history.

<table>
<thead>
<tr>
<th></th>
<th>Analysis sample (N=238)†</th>
<th>Eligible but missing surgical information</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>n (%) or median (IQR)</td>
</tr>
<tr>
<td>Age at survey (yrs)</td>
<td>238</td>
<td>5.5 (5.4-5.7)</td>
</tr>
<tr>
<td>Sex (male)</td>
<td>238</td>
<td>158 (66%)</td>
</tr>
<tr>
<td>Median deprivation score (percentile)</td>
<td>213</td>
<td>40 (16-66)</td>
</tr>
<tr>
<td>Good dentoalveolar relationship (score≤2)</td>
<td>181</td>
<td>94 (52%)</td>
</tr>
<tr>
<td>Poor dentoalveolar relationship (score≥4)</td>
<td>181</td>
<td>35 (19%)</td>
</tr>
</tbody>
</table>

†These children had at least some information about their surgery and so were included in at least one of the analysis reported in this paper.

* A test of the difference between those included in the analysis and those eligible but with no information on surgery. Chi-squared tests were used for categorical variables and t-tests for continuous variables.
<table>
<thead>
<tr>
<th>Sequence</th>
<th>N</th>
<th>(%)</th>
<th>Median (IQR)</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>157</td>
<td>70</td>
<td>3.7 (3.2-4.3)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>8.4 (6.8-9.6)</td>
</tr>
<tr>
<td>B</td>
<td>43</td>
<td>19</td>
<td>3.4 (3.2-3.9)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>8.0 (7.0-8.8)</td>
</tr>
<tr>
<td>C</td>
<td>10</td>
<td>4.4</td>
<td>4.4 (3.8-5.0)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>11.8 (11.4-12.8)</td>
</tr>
<tr>
<td>D</td>
<td>6</td>
<td>2.7</td>
<td>2.5 (1.9-3.2)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>12.7 (8.1-12.8)</td>
</tr>
<tr>
<td>E</td>
<td>3</td>
<td>1.3</td>
<td>3.8 (3.5-4.8)</td>
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<tr>
<td></td>
<td></td>
<td></td>
<td>13.1 (7.6-16.0)</td>
</tr>
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<td></td>
<td></td>
<td></td>
<td>43.5 (26.6-46.1)</td>
</tr>
<tr>
<td>F</td>
<td>1</td>
<td>0.4</td>
<td>8.2 (8.2-8.2)</td>
</tr>
<tr>
<td>G</td>
<td>0</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>H</td>
<td>0</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>I†</td>
<td>4</td>
<td>2.2</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>224</td>
<td>100</td>
<td></td>
</tr>
</tbody>
</table>

† Deprivation: lower percentiles indicate more deprived areas.

Table 3. Overall average age of surgery and between surgeon variation (Variance partition coefficient – VPC) for lip, hard palate and soft palate surgery as estimated by random intercept models.

<table>
<thead>
<tr>
<th></th>
<th>Unadjusted</th>
<th>Adjusted for surgical sequence</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Overall average age (months)</td>
<td>VPC</td>
</tr>
<tr>
<td></td>
<td>Mean (95% CI) % (95% CI) p</td>
<td>Mean (95% CI) % (95% CI) P</td>
</tr>
<tr>
<td>Lip Surgery</td>
<td>3.9 (3.7, 4.1) 15 (6, 34) &lt;0.001</td>
<td>3.9 (3.7, 4.1) 17 (7, 36) &lt;0.001</td>
</tr>
<tr>
<td>Hard Palate</td>
<td>8.7 (6.8, 10.6) 68 (53, 80) &lt;0.001</td>
<td>8.3 (7.5, 9.0) 31 (14, 56) &lt;0.001</td>
</tr>
<tr>
<td>Soft Palate</td>
<td>8.7 (8.0, 9.3) 26 (12, 46) &lt;0.001</td>
<td>8.6 (8.0, 9.2) 26 (13, 45) &lt;0.001</td>
</tr>
<tr>
<td>Hard Palate</td>
<td>7.7 (6.9, 8.5) 27 (13, 48) &lt;0.001</td>
<td>7.7 (6.9, 8.4) 18 (7, 40) &lt;0.001</td>
</tr>
</tbody>
</table>

†surgeon number 12 in plot B, figure 3 was removed.
Figure Legends:

Figure 1. Number (n) of cleft procedures by consultant surgeon and type of operation.

Figure 2. Number and variation in surgical sequence by consultant surgeon.

Figure 3. A B C D: Variation in timing (age of child at surgery) of surgery across surgeons both unadjusted and adjusted for sequence in (a) Lip Surgery (b) Hard palate repair and (c) Soft palate repair, and finally (d) Hard palate after the removal of surgeon number 12. Means and confidence intervals for each surgeon were estimated using random intercept models.