

Study models of 5 year old children as predictors of surgical outcome in unilateral cleft lip and palate

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SUMMARY This study examined features of dental occlusion in patients born with a unilateral cleft lip and palate (UCLP). The intention was to develop a 'Goslon type' index for 5 year old children. The Goslon ranking system was used on longitudinal study models taken at 5 and 10 years of age of the same patients. All patients had UCLP and this had been repaired using a Millard type lip repair and a Veau Wardill or Von Langenbeck palatal closure. There was good intra-examiner agreement for ascribing 5 and 10 year old models to one of five categories (excellent–very poor). Inter-examiner agreement on both sets of models was at worst moderate. Two of the examiners identified up to 93 per cent of 5 year old models which either remained in the same category or deteriorated by 10 years of age. At worse the results demonstrated 70 per cent of cases at 5 years of age remained in the same category or deteriorated by 10 years of age. Consensus agreement has produced five categories of outcome for these 5 year old models. This new index is to be subjected to further validation. This study has therefore provided, for the first time, a mechanism for assessing the results of CLP surgery earlier than indices already available.

Introduction

There is at present a surge of interest in the development of strategies to improve the outcome of treatment for patients born with a cleft lip and palate (Roberts *et al.*, 1991). It has become clear that poorly performed primary surgery is likely to compromise facial growth and dental development (Mars *et al.*, 1992). It is also evident that a wide range of surgical techniques exist to correct this anomaly but with no clear-cut guidelines for optimal timing or method. As a result, when outcome with one technique appears disappointing, surgeons are likely to make modifications to, or radical departures from, their current regimes. These changes are often made with little data or rationale. Until recently it has been perceived that the effects of primary surgery on the maxilla and facial growth cannot be determined until facial development is complete, usually in late adolescence. However, a large European study involving six centres demonstrated that it is possible to detect differences in outcome as early as 10 years of age (Shaw *et al.*, 1992). The ability

to predict the outcome even earlier would provide surgeons with a sound basis on which to judge their results and a rational indication that modification of a technique was justified. Recently, some evidence from a study of radiographs suggested that soft-tissue outline may be a useful indicator to evaluate quality of treatment (Mackay *et al.*, 1994). A less invasive approach would be to examine dental occlusion from dental study casts. Using this methodology, evidence is presented that treatment outcome can be detected in patients with unilateral cleft lip and palate (UCLP) at 5 years of age.

Subjects and method

Dental study casts were retrieved from the archive at Frenchay Hospital, Bristol, UK. The criteria for selection were as follows:

1. The subject was born with UCLP.
2. Primary repair of UCLP had been undertaken within Frenchay Hospital.

3. No orthodontic treatment had been carried out.
4. No secondary alveolar bone grafting had been performed.
5. Study casts were available for the subject at 5 and 10 years of age.

In all, satisfactory records for 27 subjects (17 males and 10 females) were obtained. All had a Millard type lip repair and palatal closure with either the Veau Wardill or Von Langenbeck technique.

The 54 sets of dental study casts were duplicated in white stone and the bases trimmed to standardized angles and heights.

The dental arch relationships of both the 5 and 10 year old models were assessed using the Goslon Yardstick (Mars *et al.*, 1987). In all, four examiners (two consultants, C and D, and two registrars, A and B) performed the assessment twice in one day. The second rating was made after randomly reallocating the position of the study casts to minimize possible influence of memory. Intra- and inter-examiner agreement was determined with the weighted Kappa statistic (Altman, 1991). Descriptive statistics were used to illustrate the relationship between the 5 and 10 year old Goslon scores.

Results

The mean age of the 5 year old records was 5.3 SD 0.4 years (range 5–6.4 years) and for the 10 year olds, 9.9 SD 0.8 years (range 8.3–12 years).

Intra-examiner agreement for the two separate Goslon rankings of 5 and 10 year old records is shown in Table 1. It is evident that for both the 5 and 10 year old records, agreement was good and at a similar level.

When the inter-examiner correlation for both 5 and 10 year old records was examined (Table 2), agreement was at worst moderate and at best very good (Altman, 1991).

The relationships between the 5 and 10 year old Goslon scores were then compared. Since the Goslon Yardstick was developed for 10 year olds, it is not entirely appropriate to examine agreement statistics between the Yardstick and the 5 year old records. A more representative

Table 1 Intra-examiner agreement after two separate Goslon rankings of 5 and 10 year olds models. A Kappa value of 0.8 represents very good agreement, 0.6 equates to good agreement.

| Examiner | Kappa value | |
|----------|-------------|----------|
| | 5 years | 10 years |
| A | 0.8 | 0.8 |
| B | 0.6 | 0.8 |
| C | 0.7 | 0.8 |
| D | 0.8 | 0.6 |

Table 2 Inter-examiner agreement was determined using the Kappa statistic. A value above 0.4 suggests moderate agreement, 0.6 and above, good to very good agreement. Data is representative of two rating exercises.

| 5 year models | | | | |
|----------------|-----|-----|-----|---|
| Examiner | A | B | C | D |
| A | – | | | |
| B | 0.6 | – | | |
| C | 0.5 | 0.6 | – | |
| D | 0.5 | 0.5 | 0.7 | – |
| 10 year models | | | | |
| Examiner | A | B | C | D |
| A | – | | | |
| B | 0.6 | – | | |
| C | 0.8 | 0.6 | – | |
| D | 0.8 | 0.4 | 0.7 | – |

Table 3 Relationship of Goslon rankings in 5 and 10 year old models. Data is expressed as a percentage of cases which remained the same or deteriorated.

| Examiner | 1st scoring % same/worse | 2nd scoring % same/worse |
|----------|-----------------------------|-----------------------------|
| A | 89 | 89 |
| B | 85 | 93 |
| C | 70 | 78 |
| D | 82 | 93 |

evaluation of this relationship is to compare the number of records which either retained their Goslon score or become worse from 5 to 10 years of age (Table 3). It was found firstly that in two scoring exercises on both 5 and 10 year old

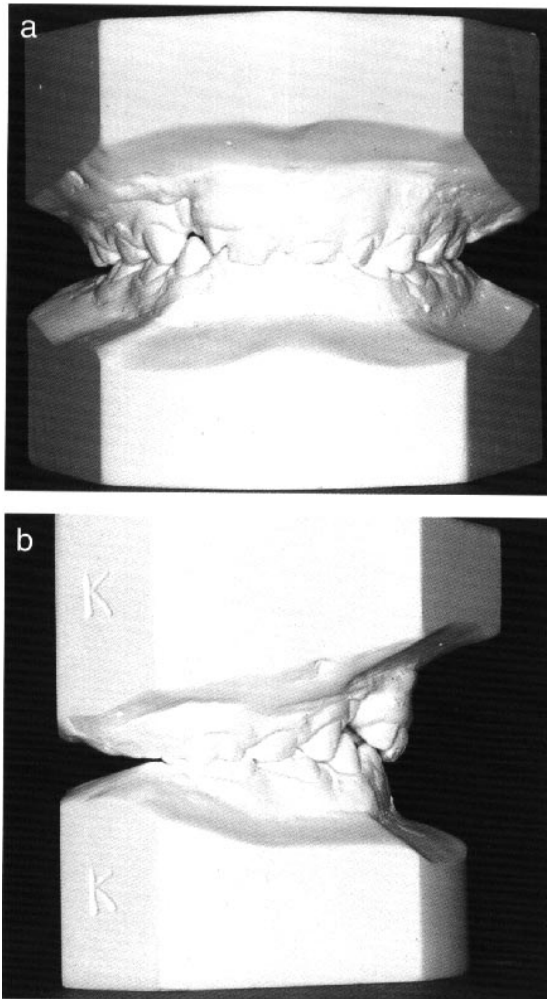


Figure 1 Representative case, (a) antero-posterior and (b) lateral view, from Group 1. This is judged an excellent result.

models, the observers were all reasonably consistent. Secondly, for two examiners (B and D), 93 per cent of the scores they made on 5 year old models stayed the same or became worse in the 10 year old models. The weakest prediction for this was 70 per cent (examiner C).

Finally from the 27, 5-year-old records, those cases where agreement was high between the examiners were selected. Two representative cases for each of the five groups were then chosen to represent a range for each category from excellent (Group 1) to very poor (Group 5). The format of the original Goslon Yardstick was retained:

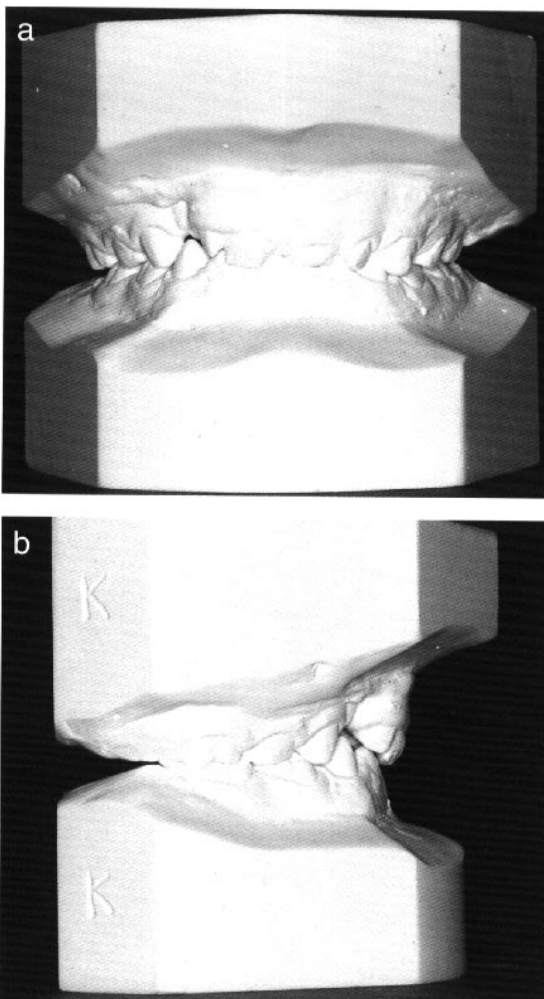


Figure 2 Representative case, (a) antero-posterior and (b) lateral view, from Group 2. This is judged a good result.

| | |
|---------|-----------|
| Group 1 | Excellent |
| Group 2 | Good |
| Group 3 | Fair |
| Group 4 | Poor |
| Group 5 | Very poor |

Examples of each category are shown in antero-posterior and lateral views in Figures 1–5.

Discussion

The aim of this study was to develop a robust and reliable index for dental relationships of 5 year old patients born with a UCLP. It is

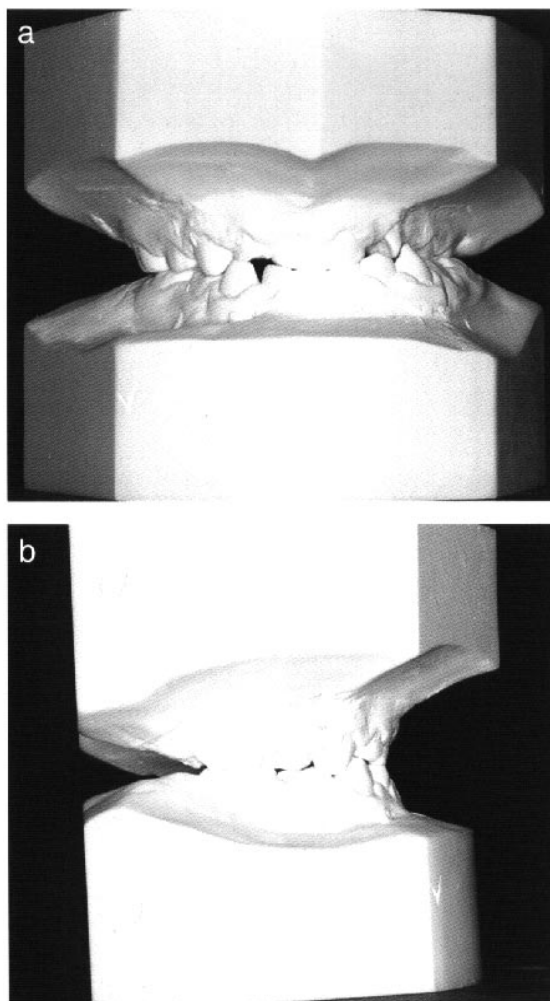


Figure 3 Representative case, (a) antero-posterior and (b) lateral view, from Group 3. This is judged a fair result.

emphasized that the sample was longitudinal and that none of the records from the 5 or 10 year old groups had received any orthodontic treatment or secondary alveolar bone grafting. The position of the teeth was therefore not influenced by these variables. At the time, the standard procedures for children born with UCLP and referred to Frenchay Hospital was to repair the lip at 3 months of age and the palate at 6 months. Palatopharyngoplasty was performed in cases with speech problems and in all there were five such patients in the sample. There is no definitive proof that this latter procedure can influence facial growth.

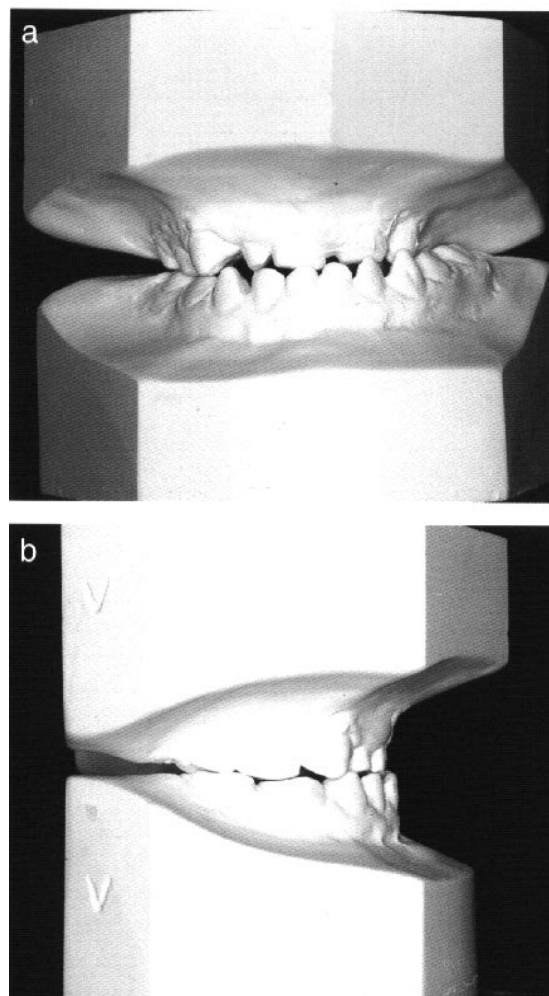


Figure 4 Representative case, (a) antero-posterior and (b) lateral view, from Group 4. This is judged a poor result.

The development of early predictors of outcome in cleft care is timely and relevant. The need for these markers is urgent in those countries where standards of care are perceived to be below the best European centres (Shaw *et al.*, 1992). The problem of poor outcome is compounded by surgeons who perform few operations each year (Williams *et al.*, 1994). Small caseloads make meaningful statistical analysis almost impossible for an individual surgeon. It has been suggested that larger centres could act as a reference norm against which smaller units could measure their success (Ward Booth, 1995). The index that has been described

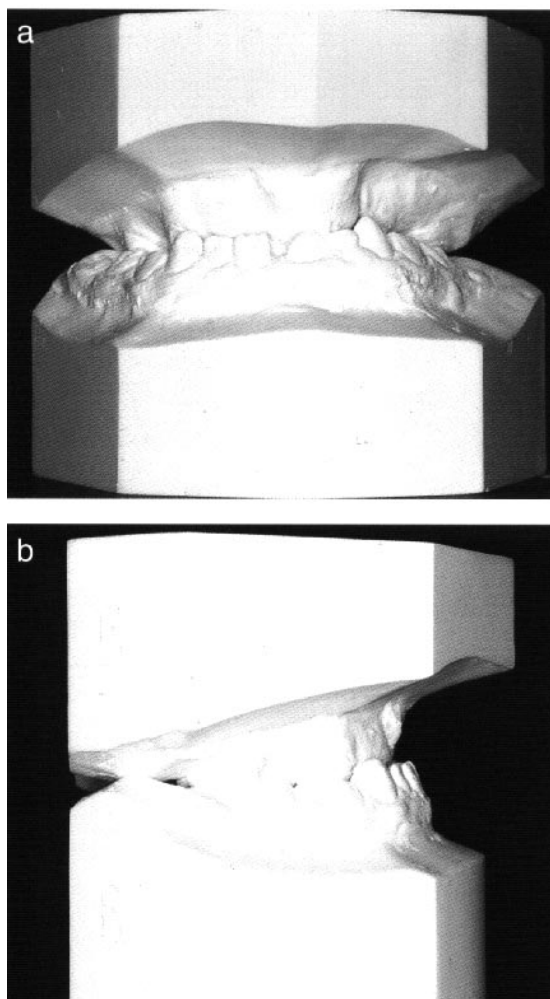


Figure 5 Representative case, (a) antero-posterior and (b) lateral view, from Group 5. This is judged a very poor result.

in this report could be used for this comparison and the percentiles of the five groups in the total sample would be an important parameter in the assessment of surgical outcome. However, it could take many years of surgery before poor results could be confirmed on a sound statistical basis even if this period saw only one operator in post.

Attempts at estimating sample sizes suggest that a surgeon receiving 40 new referrals of all cleft types each year would reach a point of meaningful audit some 8.5 years after appointment. This calculation assumes differences are

detectable at 5 years of age (Shaw *et al.*, 1992), but as the majority of cleft surgeons in the U.K. perform less than 10 repairs of the lip and palate each year (Williams *et al.*, 1994), the audit of outcome is impracticable and poor results are likely to remain undetected. The development of a rating system to examine surgical outcome at the age of 5 years we believe represents a significant step in the treatment of patients with a cleft lip and palate. The advantage of this index over the existing 10 year old Goslon Yardstick is that the 5 year old child is 'clean', with respect to surgical procedures, and not 'contaminated' by other treatments such as secondary alveolar bone grafting or orthodontics. With the move towards earlier alveolar bone grafting to allow the eruption of permanent teeth (lateral incisors and canines) it is unlikely that future 10 year old samples will have all subjects similarly matched as far as treatment received is concerned. Finally, obtaining records at the age of 5 is coincident with the recommendations of the International Committee on Cleft Documentation and Measurements (Lee *et al.*, 1993) and should enable inter-centre studies to be undertaken relatively rapidly. This index is now being subjected to validation against a large sample of patients with UCLP.

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