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Scientific Section

Early Detection of Differences in Surgical Outcome for Cleft Lip and Palate

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Abstract: *This study examined the dento-alveolar relationships of 5-year-old children born with a unilateral cleft lip and palate with primary surgical repair performed in one of two centres (Bristol or Oslo). The Bristol sample comprised 46 sets of study models and the Oslo CLP Growth Archive provided 54 cases with a very similar sex distribution. We used a recently developed 5-year-old index to measure differences in outcome between the two centres. The Oslo sample were assessed as having up to 57 per cent in the ideal groupings (1 and 2), in the Bristol group this was only 35 per cent. Bristol had up to 46 per cent of cases assessed in the worst groups (4 and 5). The comparative figure from the Oslo group was 15 per cent. These results suggest that it is possible to detect differences in surgical outcome at 5 years of age.*

Index Words: Early Surgical Outcomes, Inter-centre Comparison Unilateral Cleft Lip and Palate.

Introduction

There is currently an intense interest in the early detection of outcomes in subjects with cleft lip and palate. Since this anomaly requires multi-disciplinary care, the establishment of outcomes for a number of aspects is an area of much activity. It is broadly agreed that mid-facial growth is probably a reasonable indicator of surgical outcome, since it appears that primary surgery used for the correction of the lip and palate defect has some effect on the maxilla (Semb, 1991). This, in turn, may be reflected in the dental arch relationships and this forms the basis for the development of indices using dental study casts (Mars *et al*., 1987). It was thought that the earliest that differences in outcome could be detected from study models was 10 years of age (Mars *et al*., 1987, 1992). Noverraz *et al*., (1993) however, provided some evidence to suggest that the Goslon Yardstick may be a useful method for longitudinal assessment throughout the development of the occlusion. Using similar methodology an Index for assessing outcome at the age of 5 years has been proposed (Atack *et al*., 1997a).

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The development of early predictors of outcome is both timely and relevant since it is not known what the optimum technique is for repair of cleft lip and palate (Cabre *et al*., 1995; Markus and Ward-Booth, 1995a,b; Piggott, 1995; Sell, 1995; Timmons, 1995). There are many different surgical operations described (Mølsted, 1992) and no agreement on the timing or technique which produces the best result. Furthermore, despite strong indications, there is currently debate on whether this surgery should be undertaken in isolated or centralised units. Whatever arguments are in place for the organization of cleft care it cannot be disputed that in order to show differences in outcomes between operators or techniques there have to be sufficient cases to provide statistical comparisons between groups of patients. All cleft palate carers agree, whatever their beliefs on best care, that the continual assessment and auditing of results are essential if the management of cleft lip and palate patients is to be advanced. If it is possible to detect differences in the outcome of cleft care in children at 5 years of age, it has been calculated that on a case load of 30 new cases per year, it would take nearly 9 years for the operator or unit to provide sufficient cases for meaningful comparison with other centres (Shaw *et al*., 1996). Some evidence has been presented that using soft tissue form as a measure it is possible to detect differences in outcome

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between centres at 5 years of age (Mackay *et al*., 1994). Apart from this there is scant evidence of objective measures for early cleft outcomes.

The purpose of this study was to examine whether differences in dental arch relationships of 5-year-old children born with a unilateral cleft lip and palate (CLP) could be detected.

Materials and Methods

Study models of 5-year-old subjects with repaired unilateral CLP were obtained from the archives of two surgical units, Bristol and Oslo.

Sample 1 (Bristol)

Forty-six sets of study models were retrieved from the model collection at Frenchay Hospital. No syndromic cases were included. All subjects had their primary surgical repairs undertaken at Frenchay Hospital. A variety of surgical techniques had been used and multiple operators had undertaken these procedures. In general, the lip repair was performed at three months of age whereas the palatal repair was undertaken between the age of 6 months to 1 year.

Sample 2 (Oslo)

Fifty-four sets of study models were obtained from the Oslo CLP Growth Archive. All these subjects had their repairs undertaken by a single surgeon. A Millard lip repair was performed at 3 months together with a vomerine flap for nasal layer closure. The palate was repaired at 18 months with a Von Langenbeck closure.

No primary bone grafting was undertaken in either unit. Samples from both units were complete UCLP, they were not consecutive or independently vetted. The Frenchay cases were not preselected in that all available models were obtained. A similar sized group operated on at a similar time were retrieved from the Oslo CLP growth archive.

The dental arch relationships of the study models were assessed using the 5-year-olds' Index (Atack *et al.*, 1997a,b), a means of subjectively categorizing arch relationships of CLP children in terms of A–P, vertical and transverse discrepancies in the deciduous dentition using reference study models. Each sample was assessed twice in one day by two examiners: one orthodontic consultant (A) and one registrar (B). A suitable period of time (1 week) was allowed between the two scorings. The position of the study models was reallocated using randomly generated numbers for the second scoring to minimise the possible influence of memory on the results.

The intra- and inter-examiner agreement was determined using the weighted kappa (κ) statistic which takes into account the degree of disagreement within the results (Table 1). Percentages were used to illustrate the differences between surgical units. Statistical calculations were undertaken using Survey Plus, Survey Analysis-Statistics, Version 4·50 (Providence Software Services).

Results

Sample 1 comprised 15 females (33 per cent) and 31 males (67 per cent). Sample 2 comprised 23 (38 per cent) females and 37 (62 per cent) males. All models from both samples were at a similar stage of dental development.

The weighted κ values demonstrated that there was moderate to very good agreement both within and between examiners when assessing both samples. The intra-examiner agreement is shown in Table 2. The inter-examiner agreement is shown in Table 3. The 5-year-olds' Index was found to be reproducible and this supports earlier work (Atack *et al*., 1997b).

The percentiles within each Index category for the Bristol and Oslo samples are illustrated in Tables 4 and 5. Graphic representation of these results are illustrated in Figures 1 and 2.

TABLE 1 *Level of agreement: weighted κ values (taken from Altman 1991)*

Value of κ	Strength of agreement
0·20 $\geqslant$	Poor
0·21–0·40	Fair
0·41–0·60	Moderate
0·61–0·80	Good
0·81–1·00	Very Good

TABLE 2 *Intra-examiner agreement (weighted κ values). The lower confidence limits are in parenthesis*

Examiner	Sample 1 (Bristol)	Sample 2 (Oslo)
A	0·76 (0·62)	0·86 (0·76)
B	0·87 (0·75)	0·96 (0·90)

TABLE 3 *Inter-examiner agreement (weighted κ values). The lower confidence limits are in parenthesis*

Examiner	Sample 1 (Bristol)	Sample 2 (Oslo)
First assessment	0·76 (0·62)	0·50 (0·34)
Second assessment	0·65 (0·49)	0·56 (0·40)

TABLE 4 *The results of two assessments of the Bristol Sample. The percentage of models within each category is shown. The absolute number is given in parenthesis*

Index category	Examiner			
	A		B	
	First scoring	Second scoring	First scoring	Second scoring
1	11 (5)	15 (7)	13 (6)	13 (6)
2	22 (10)	13 (6)	17 (8)	22 (10)
3	24 (11)	26 (12)	26 (12)	24 (11)
4	24 (11)	28 (13)	24 (11)	22 (10)
5	20 (9)	18 (8)	20 (9)	20 (9)

TABLE 5 *The results of two assessments of the Oslo Sample. The percentage of models within each category is shown. The absolute number is given in parenthesis*

Index category	Examiner			
	A		B	
	First scoring	Second scoring	First scoring	Second scoring
1	11 (6)	15 (8)	31 (17)	28 (15)
2	37 (20)	37 (20)	26 (14)	29 (16)
3	37 (20)	33 (18)	28 (15)	28 (15)
4	11 (6)	11 (6)	9 (5)	9 (5)
5	4 (2)	4 (2)	6 (3)	6 (3)

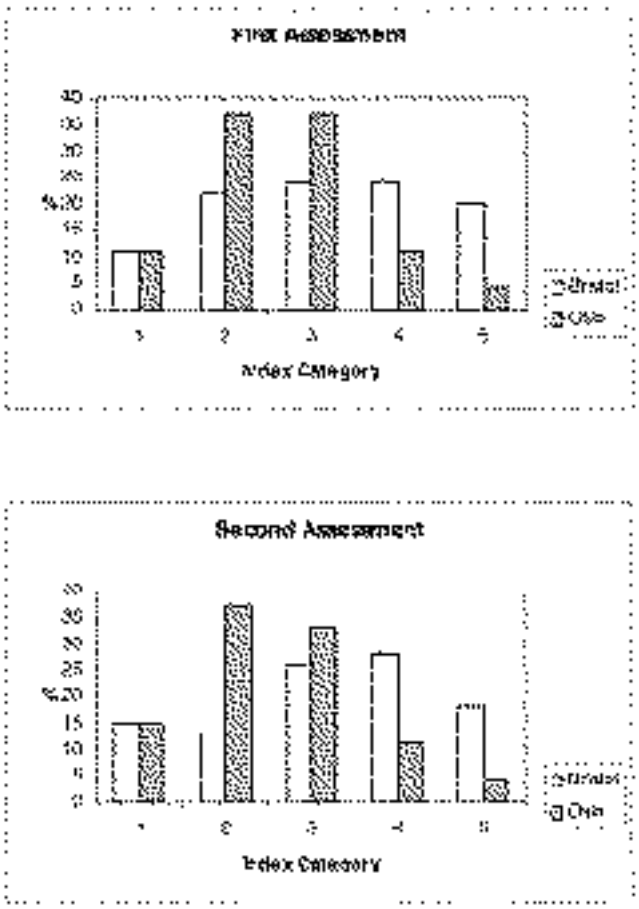

FIG. 1 Examiner A: graphic representation of the percentiles in each Index group for the first and second assessments.

It is widely agreed that those cases ascribed to groups 4 and 5 are poor surgical results which may require orthognathic surgery during late adolescence, whereas those assessed in groups 1 and 2 are considered to have good long-term growth outcomes. The results showed that the Oslo sample had a far greater number of subjects assessed in groups 1 and 2 compared to the Bristol sample. In contrast, the Bristol sample had a far greater percentage of subjects in groups 4 and 5 compared to the Oslo sample. These results suggest that it is possible to detect differences in surgical outcome at 5 years of age.

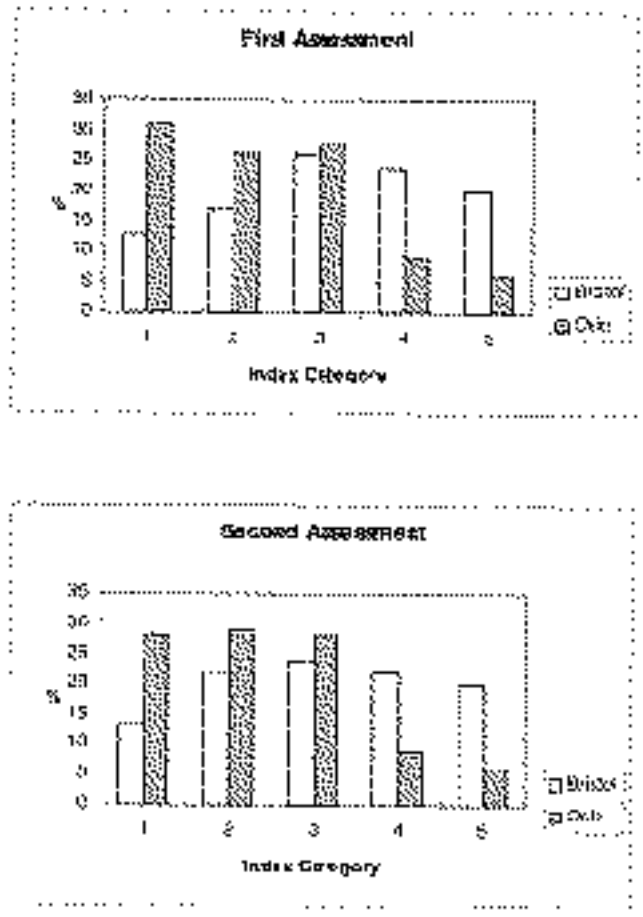

FIG. 2 Examiner B: graphic representation of the percentiles in each Index group for the first and second assessments.

Discussion

This study appears to show that differences in outcome between two centres are apparent at 5 years of age. This is important as previously the earliest these differences had been detected was in 10-year-olds with the Goslon Yardstick or variations on this system (Friede *et al*., 1991; Mars *et al*., 1987, 1992). Dental arch relationships measured with the Goslon Yardstick were an important and sensitive outcome in the Eurocleft Study (Mars *et al*., 1992; Asher-McDade *et al.*, 1992; Mølsted *et al.*, 1992; Shaw *et al*., 1992a,b). It became clear that using this system, differences in dental arch relationships could be detected between the six centres and that the two United Kingdom centres compared unfavourably to the other European Centres. Indeed, the number of patients likely to require orthognathic surgery (those categorized in groups 4 and 5) was 50 per cent in the worst centre, an order of magnitude greater than the best centre (5 per cent). Further work has corroborated these findings. Hathorn *et al.* (1996) examined the dental study casts of 10-year-old subjects with unilateral CLP from Bristol and Oslo. The dental arch relationships were measured with the Goslon Yardstick and they found a very similar pattern of categorisation for their cases (14 per cent in groups 1 and 2, 55 per cent in groups 4 and 5). Not surprisingly the records of the 5-year-

olds measured with the 5-year-olds' Index and reported here were similar. There was however a tendency to score the 5-year-olds slightly more favourably with 42–46 per cent in groups 4 and 5. Bearing in mind that it appears that maxillary growth worsens with age in patients with unilateral CLP compared to a normal population (Semb, 1991), the percentage of the 5-year-olds in groups 4 and 5 is likely to be higher at ten years of age.

Cephalometric radiographs have been suggested as a means of assessing outcome, but these are of limited use, particularly as an invasive investigation in patients with abnormal anatomy. Some cephalometric landmarks may be difficult to identify in subjects with CLP because of the distortion of maxillary skeletal structures (Mølsted *et al*., 1992). High image quality can reduce these problems but there is often difficulty in identifying the form of the maxilla and therefore in determining its antero-posterior position (Mackay *et al*., 1994). In the Eurocleft Study the results of cephalometric analysis were disappointing, principally because of the difficulty in identifying landmarks and in standardising radiographic equipment in different centres (Shaw *et al.*, 1992b). The use of study casts, which is a minimally invasive investigation, would seem to offer great scope as an early outcome measure in subjects with CLP.

How then can the differences in outcome between the two units be explained? To a large extent the differences have already been explored in the Eurocleft Study where it seems that units where case loads by an individual surgeon are low may perform suboptimal surgery (Roberts *et al*., 1991; Mølsted, 1992; Shaw *et al*., 1992b). The practice of low volume operators appears widespread in England and Wales with fewer than six surgeons having a case load of 30 patients with CLP in all forms in 1 year (Williams *et al*., 1994). If this practice persists it will remain difficult to identify units, surgeons, and techniques which produce poor results until the end of a surgeon's career. The data produced reinforces the view of the Eurocleft Study that better surgical outcomes are achieved by high volume operators and these appear detectable at 5 years of age. Clearly, as outcomes for speech, hearing, appearance, and psychosocial well-being are developed it will be important to integrate all facets to determine quality of cleft care. Moreover, the earlier these measures can be detected the sooner rational changes to protocols can be made where the quality of outcome is poor.

There are two other differences between the two units. The methods of repair in the Bristol unit varied and the surgical notes were unable to yield accurate information on the exact nature of the surgical method. For this reason it was unfair to ascribe differences to a specific technique. Furthermore, the multiple operators using different techniques in Bristol precluded analysis because of the small numbers in the different groups. A further variable was the timing, the palate in some of the Bristol patients was repaired at a younger age than the Oslo patients. In the latter group the timing was consistent, in the former, variability in the staging of this operation resulted in a number of small groups on which statistical analysis was not possible. We felt that the lack of surgical timing and consistency of surgical method meant that these groups were unable to provide meaningful information on these two aspects.

Conclusions

1. Differences in dental arch relationships in patients with unilateral CLP are apparent at 5 years of age.
2. The use of an index for these 5-year-old dental study casts appears to show differences in outcome between two centres.
3. With correct study design it may be feasible to compare outcomes from techniques and individual surgeons.

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