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Evaluation of primary surgical outcomes in New Zealand patients with unilateral clefts of the lip and palate

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Objective: To evaluate and compare the primary surgical outcomes of complete unilateral cleft lip and palate (UCLP) patients in two New Zealand cleft care centres.

Methods: This is a retrospective study of two providers of cleft care in New Zealand: Centre A in the North Island and Centre B in the South Island of New Zealand. Pre-orthodontic study models were evaluated from 28 UCLP patients from Centre A with primary surgical repairs performed between 1987–1999 and 31 UCLP patients from Centre B with primary surgical repairs performed between 1984–2000. Dental arch relationships were measured using the Goslon Yardstick. A Goslon score of 1 is considered to be an excellent outcome, whereas a score of 5 is a very poor treatment outcome.

Results: Intra- (Kappa: 0.84 – 0.93) and inter-examiner (Kappa: 0.63 – 0.69) reliabilities revealed good to very good agreement between examiners using the Goslon Yardstick. The mean Goslon score for Centre A was 3.5, with no cases in Group 1, five cases in Group 2 (17.9 per cent), nine cases in Group 3 (32.1 per cent), 11 cases in Group 4 (39.3 per cent) and three cases in Group 5 (10.7 per cent). The mean score for Centre B was 3.1, with one case in Group 1 (3.2 per cent), nine cases in Group 2 (29.0 per cent), eight cases in Group 3 (25.8 per cent), 11 cases in Group 4 (35.5 per cent) and two cases in Group 5 (6.5 per cent). There were no statistically significant differences between the two centres ($p > 0.05$).

Conclusions: The outcome scores from the two cleft centres, based on historic records, were disappointing and higher than expected. It is recommended that a review of primary surgical protocols be implemented to ensure outcomes comparable with international standards. The results provide useful benchmarks for future comparisons of treatment.

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Introduction

It is important that treatment outcomes of patients who present with cleft lip and palate are regularly evaluated, to ensure quality and consistency of patient care, as well as allow outcome comparisons between different cleft care centres.[1–3] For clinical audits to be conducted effectively, emphasis should be placed on the development of methods of assessing treatment outcomes.[4] Previous reports have recommended indices that assess arch relationships and one suggested method is to measure the degree of crossbite for individual teeth in order to calculate an overall score.[5,6] However, this method can be cumbersome and time-consuming and inaccurately represent the overall severity of the malocclusion. It is possible that a mild generalised irregularity may yield a higher score than a more severe but localised anomaly.[4]

An effective method used in a number of multicentre audits to assess dental arch relationships in complete unilateral cleft lip and palate patients (UCLP) is the Goslon (Great Ormond Street London and Oslo) Yardstick.[7,8] Unlike other indices, the Goslon Yardstick assesses the difficulty of orthodontic correction as well as the severity of malocclusion. The measure has been shown to be simple and reliable when used by calibrated examiners.[7,9,10] It allows

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categorisation of dental arch relationships in the late mixed or early permanent dentition stage into five discrete categories. The anteroposterior relationship of the arches is of greatest clinical importance as it reflects the underlying skeletal relationship. Generalised dental crowding and irregularity is relatively less important. Accordingly, Goslon Groups 1 and 2 (excellent/good result) have occlusions that require simple orthodontic treatment or none at all, while Group 3 (fair result) requires complex orthodontic treatment to correct malocclusions, although a good result can be anticipated. In contrast, Goslon Groups 4 and 5 (poor/very poor result) have poor dental arch relationships and often require orthognathic surgery to correct underlying skeletal disharmonies.[4,7]

Previous studies have suggested a higher rate of non-syndromic orofacial clefts in New Zealand compared with European studies.[11–13] Previously reported New Zealand data has revealed an incidence of 1.5 to 1.94 per 1000 live births in patients of European descent[11–13] and 2.26 per 1000 live births in patients of Maori descent.[12] In contrast, the incidence in the United Kingdom and in most parts of Europe ranges from 1.28 to 1.81 per 1000 live births.[14,15] The relatively high incidence of non-syndromic orofacial clefts in New Zealand further illustrates the importance of auditing and assessing the outcome of primary surgery, in order to achieve sustainability of improved cleft care standards.

Treatment outcomes of cleft care in New Zealand have not been previously audited using the Goslon Yardstick. Therefore, the aim of this study was to evaluate and compare the primary surgical outcomes of complete unilateral cleft lip and palate (UCLP) patients in two New Zealand cleft care centres using this Yardstick. A secondary objective was to compare the New Zealand findings with those from the Eurocleft study[1,7] which examined primary surgical outcomes in six European centres. It is expected that the data will provide a useful baseline against which future national clinical audits may be compared.

Materials and methods

An audit of primary surgical treatment outcomes in New Zealand children with complete UCLP was carried out in June 2008. The study included two providers of cleft care identified as Centre A in the North Island and Centre B in the South Island of New Zealand. The sample consisted of 28 UCLP patients presenting consecutively for alveolar bone grafting from Centre A from 1987–1999, and 31 UCLP patients presenting consecutively for alveolar bone grafting from Centre B from 1984–2000.

All had a history of non-syndromic complete unilateral cleft lip and palate, with surgical lip repair completed approximately 3 months after birth and palate repair approximately 9 months after birth. Patients with Simonart's bands were excluded and all patients were reassessed prior to commencing orthodontic treatment.

Dental study models were obtained from the orthodontic clinics of the dental departments of the two respective hospitals, and de-identified. The models were assessed by two orthodontists experienced in cleft care and who had been calibrated in the use of the Goslon Yardstick. During assessor calibration, very good intra-examiner agreement (Weighted Kappa: 0.82, 0.89) and good inter-examiner agreement (Weighted Kappa: 0.77) was demonstrated. A set of Goslon reference models representing each Yardstick category was available for comparison and reference during the model assessment. Each assessor worked independently and repeated the model examination in order to evaluate intra-examiner reliability. Neither assessor was associated with the two cleft care centres involved in the present audit.

Data were analysed using the Statistical Package for Social Sciences (SPSS v 16.0, SPSS Incorporated, Chicago, IL, USA). Bivariate associations were investigated using Chi-squared tests with the alpha level set to 0.05. Intra-examiner and inter-examiner reliabilities were calculated using weighted Kappa statistics.[16] A Kappa statistic greater than 0.6 indicated good agreement, while one greater than 0.8 indicated very good agreement.[17]

Results

The mean age of the sample was 9.9 years (S.D: 1.53) in Centre A and 9.6 years (SD: 1.69) in Centre B. There were greater proportions of males and left-sided UCLP patients in both centres (Table I). No statistically significant differences in patient demographics were found between the two centres.

The intra-examiner reliability of both examiners was very good, as indicated by the Weighted Kappa in the Centre A (Examiner A: 0.93; Examiner B: 0.84)

Table I. Sociodemographic characteristics and distribution of UCLP location in the New Zealand study samples.

	NZA N (Per cent)	NZB N (Per cent)	Total N (Per cent)
Gender			
Male	16 (57.1)	22 (71.0)	38 (64.4)
Female	12 (42.9)	9 (29.0)	21 (35.6)
Age			
7–10 years	19 (67.9)	23 (74.2)	42 (71.2)
11–14 years	9 (32.1)	8 (25.8)	17 (28.8)
Location			
Left	16 (57.1)	19 (61.3)	35 (59.3)
Right	12 (42.9)	12 (38.7)	24 (40.7)

Table II. Goslon Yardstick distribution in the New Zealand study samples.

Golson grade	NZA N (Per cent)	NZB N (Per cent)	Total N (Per cent)
Group 1	0 (0.0)	1 (3.2)	1 (1.7)
Group 2	5 (17.9)	9 (29.0)	15 (25.4)
Group 3	9 (32.1)	8 (25.8)	16 (27.1)
Group 4	11 (39.3)	11 (35.5)	21 (35.6)
Group 5	3 (10.7)	2 (6.5)	6 (10.2)

and Centre B samples (Examiner A: 0.91; Examiner B: 0.86). The inter-examiner reliability was also considered good (Weighted Kappa: > 0.6) in both centres. The distribution of Goslon scores for the New Zealand centres is shown in Table II. The mean Golson score for Centres A and B were 3.5 and 3.1 respectively. There were no statistically significant differences in the Goslon score between the two centres. Centre A and Centre B performed similarly to the worst ranked of the European centres reported by the Eurocleft study[1,7] (Figure I). Nearly half of the Centre A and Centre B samples were classified as having poor/very poor (Groups 4 and 5) treatment outcomes.

Discussion

The Goslon Yardstick was used to evaluate dental arch relationships and primary surgical treatment outcomes of complete UCLP in two New Zealand cleft care centres. This Yardstick has been widely used and shown to be a reliable and robust means of measurement when used in multicentre audits.[1,4,8–10,18–20] Although subjective,[4] it is a rapid and simple way of measuring dental arch relationships. It has the benefit of being based on the clinical features that are most difficult to treat, thus giving a practical indication of the requirements of treatment, whether by orthodontics or a combination of orthodontics and surgery.[7]

The Eurocleft study[1,7] involved the direct Goslon Yardstick comparison of treatment outcomes from six European treatment centres. It showed that centralised and standardised treatment protocols, in cleft centres that managed a large number of clefts, were associated with the best treatment results. A comparison of the Goslon ratings between the Eurocleft study and the New Zealand UCLP audit indicates that New Zealand patients would benefit from improvements in primary cleft care. The difference in the standard of treatment outcomes between the New Zealand sample and those of the best Eurocleft centres appears to be considerable. For example, the proportion of patients with poor treatment outcomes (Groups 4 and 5) in the present sample (46 per cent) was most comparable with the centre which had the worst treatment outcome of the European centres. By comparison, the three best centres in the Eurocleft study had fewer than 10 per cent of its samples in Groups 4 or 5.[7] The present study's findings suggest that a large proportion of the New Zealand patients are likely to have underlying skeletal discrepancies that may require future orthognathic surgery.

There were no statistically significant differences in treatment outcome between the two cleft care centres, even though different surgical techniques for palatal repair were preferred by each centre. The preference in Centre A was for the Wardill-Kilner procedure, which was performed by a number of surgeons. Centre B had one operating surgeon who preferred the Von Langenbeck procedure. Ethnic data for this sample was not recorded and any racial variations within the sample, which may affect the Golson ratings, were not considered.

Overall, the audit results were disappointing. A possible reason to explain the unsatisfactory treatment outcomes observed in New Zealand is the relatively low population density and the low number of sur-

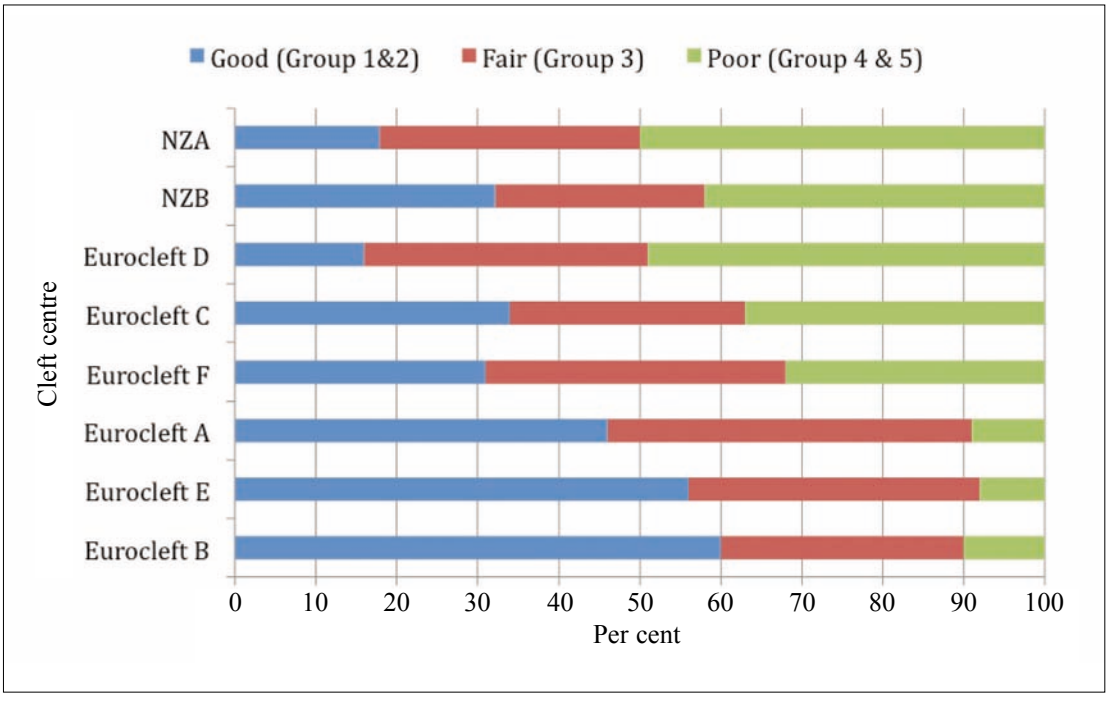

Figure 1. Comparisons of Golson score distributions between the present study and the Eurocleft study.[7]

gical patients. At present, there are five treatment centres throughout New Zealand, with varying numbers of cleft patients. To centralise the New Zealand treatment model further would mean increasing travel distances for patients which adds to the burden of care associated with UCLP. However, a centralised model would help increase operator volume, which has been suggested as an important factor in treatment proficiency and outcome.[2,19]

It is also worthwhile noting that the collected data is historical and the audited cleft centres have moved to a model in which all primary surgery is performed by a principal surgeon in each centre. The effects of such a model on primary surgical outcomes will be evaluated at a later date in an expanded plan to assess all cleft care centres in New Zealand.

Although the present audit used the Goslon Yardstick, it is noteworthy that UCLP evaluation involves other indices. Factors such as hearing, speech and facial profile, as well as self-reported quality of life, are additional parameters which must be taken into account when assessing overall treatment outcome. Improvements in skeletal base relationships should not come at the expense of any of the other parameters.

A Clinical Standards Advisory Group report[2] advised that for a high standard of cleft care to be achieved, a national comprehensive database is needed. A New Zealand database was established in 2000 to ensure uniform data collection for all cleft patients. This data was unavailable for the current study. It is expected to provide a valuable resource and source of information for future clinical audits as it will contain more detailed, robust and standardised data of New Zealand cleft patients. Ethnicity data is now routinely collected which will allow for future research into the incidence and treatment of orofacial clefts in ethnic minorities.

Conclusions

The primary surgical outcomes of complete UCLP patients in New Zealand were audited in two cleft care centres using the Goslon Yardstick. There were no statistically significant differences between the two New Zealand treatment centres, even though different primary palatal repair surgical procedures were preferred. The overall treatment outcomes from New Zealand compare unfavourably with the European centres reported in the Eurocleft study. Future national clinical audits need to be carried

out to evaluate and improve the standard of cleft care.

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