The impact of orthodontic treatment on the dental occlusion and oral health-related quality of life of individuals with cleft lip and/or palate

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Background: Children with cleft lip and/or palate (CL/P) are a special group of orthodontic patients. Oral health-related quality of life (OHRQoL) is an important aspect of treatment outcome that is becoming popular in its application in orthodontics, but its relationship with clinical treatment outcomes remains unclear.

Objectives: To compare OHRQoL and clinical treatment outcomes of a sample of children with cleft lip and/or palate and non-cleft children before and after orthodontic treatment in order to determine whether there is an association between the two outcome measures.

Methods: A prospective observational longitudinal study was conducted of 60 consecutive adolescent patients (33 CL/P and 27 non-cleft) who received orthodontic treatment at Christchurch Hospital, New Zealand. The two outcome measures were a self-reported OHRQoL outcome assessed by the short form 8 item Child Perception Questionnaire (CPQ-8SF) and an orthodontic treatment outcome assessed by the Peer Assessment Rating (PAR) index.

Results: Both CL/P and non-cleft groups had comparable pretreatment malocclusion severity with similar baseline and end of treatment OHRQoL. Although both groups showed similar improvements in OHRQoL following orthodontic treatment, the non-cleft group had significantly greater improvement in their PAR outcome. There was no association detected between OHRQoL and the PAR improvement.

Conclusion: Both study groups had similar OHRQoL changes following orthodontic treatment despite the non-cleft group having better orthodontic treatment outcomes. There is a lack of correlation between the patient’s self-assessment OHRQoL and clinically based orthodontic treatment outcomes.

(Aust Orthod J 2019; 35: 119-126)

Received for publication: October 2018
Accepted: March 2019

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Introduction

Cleft lip and/or palate (CL/P) is the most common congenital craniofacial anomaly, affecting 1.79 per thousand live births in New Zealand.1 CL/P is associated with a range of impairments related to feeding, speech, hearing, malocclusion, poor facial aesthetics and psychosocial issues.2 Cleft treatment, which can start in the first few weeks of life and often continues through to early adulthood, can carry a high burden of care. Co-ordinated surgical and nonsurgical treatment is provided by clinicians working within a multidisciplinary team. Orthodontic treatment plays a key role in the rehabilitation of affected patients.

The multifactorial aspects of CL/P care mean that outcomes can be difficult to assess by clinical indices alone. Oral health-related quality of life (OHRQoL) measures are considered to be a better gauge of treatment outcome.3 There are a variety of instruments for measuring OHRQoL in children,4 of which the Child Perception Questionnaire has been commonly
applied in studies including children presenting with a cleft.\textsuperscript{3-5} The original 37-item Children Perception Questionnaire (CPQ\textsubscript{11-14}) was developed specifically for children aged 11–14 years old and has been applied to children with a wide range of orofacial conditions.\textsuperscript{3} The original measure is considered long and difficult to administer in clinical settings and to facilitate its applicability, short versions of the CPQ\textsubscript{11-14} were developed, giving rise to the Impact Short Forms ISF:16 and ISF:8.\textsuperscript{6} The short-form versions of the CPQ\textsubscript{11-14} are more commonly used, due to their lower respondent burden while retaining acceptable validity and reliability.\textsuperscript{7-10} Previous New Zealand studies have identified a relationship between the CPQ\textsubscript{11-14} and malocclusion,\textsuperscript{11-13} with improvements following orthodontic treatment, but have not included children with CL/P.\textsuperscript{13} International studies using the CPQ\textsubscript{11-14} have shown the presence of CL/P has a negative influence on OHRQoL.\textsuperscript{3,5} A New Zealand study using a different OHRQoL measure investigated the impact of orthodontic treatment, including those with CL/P alongside non-cleft individuals.\textsuperscript{14} The study found that orthodontic treatment improved the OHRQoL of all patients, although a repeat study using the same patient cohort at a five-year follow-up showed deterioration. The CL/P group reported the greatest deterioration down to similar scores as their pretreatment OHRQoL status.\textsuperscript{15} However, the OHRQoL measure used in the two studies (OHIP-14) was not child-specific and the orthodontic treatment outcome measures were not assessed.

Accordingly, the present study had two objectives: (1) to compare the OHRQoL of a sample of children with cleft lip and/or palate (CL/P) and non-cleft children before and after orthodontic treatment using a child specific short form questionnaire (CPQ\textsubscript{11-14}-ISF8); and (2) to compare the clinical treatment outcomes between the two groups to determine whether there is an association between OHRQoL and clinical treatment outcomes.

**Methods**

The study sample consisted of 60 consecutive adolescent orthodontic patients who received treatment at Christchurch Hospital, New Zealand, between 2007 and 2015. Although the majority of the orthodontic patients treated at the hospital clinic are adults requiring orthognathic surgery for severe skeletal discrepancies, the eligibility of adolescents is restricted to those presenting with a craniofacial anomaly. These include patients with an orofacial cleft or those in receipt of a State benefit and presenting with a severe or handicapping malocclusion as defined by the Dental Aesthetic Index (DAI). The DAI is an epidemiological index that assesses the relative social acceptability of dental appearance by assessing 10 intraoral measurements that are weighted to produce a numerical score with a constant of 13 added to the final tally. The DAI scores are classified into minor (15–25), definite (26–31), severe (32–35) and handicapping (>36) malocclusions.\textsuperscript{16}

A common problem for all studies involving children with a cleft is the identification of adequate patient numbers.\textsuperscript{17} Despite including all children with a cleft condition who received orthodontic treatment at the hospital clinic in the present study, it required eight years to accumulate sufficient numbers (N = 33). This consideration was based on previous recommendations on the minimum number of cleft patients required to assess treatment outcomes,\textsuperscript{18,19} and has been used by centres participating in large international treatment outcome studies.\textsuperscript{17,20}

Participants were categorised as either non-cleft adolescent patients (mean age 14.7+/−1.8) with a severe or handicapping malocclusion (N = 27), or cleft adolescent patients (CL/P) (mean age 12.1+/−2.0) who received orthodontic treatment as part of their multidisciplinary cleft management (N = 33). All patients received comprehensive orthodontic treatment with fixed appliances. Patients who received other forms of orthodontic treatment or who required orthognathic surgery once facial growth had ceased were excluded. All participants were treated by two specialist orthodontists in the same facility.

The hospital’s electronic management system was interrogated to record patient demographic data. Socioeconomic status was determined using an area-based measure, the NZDep13, in which nine variables collected from the national census database were used to allocate a deprivation score to a ‘mesh block’.\textsuperscript{21} Each patient’s residential address was geocoded to its corresponding mesh block deprivation score, in which areas with scores 1–3 are classified as ‘low deprivation’, 4–7 ‘medium deprivation’ and 8–10 ‘high deprivation.’
The dental health status and treatment duration information was collected from dental charting on Titanium V3.7 program (Spark Dental Technology, Auckland). The dental health records were collected by dental therapists in their routine examinations as part of New Zealand’s Community Oral Health Service. From this information, the dental health status, as defined by the numbers of decayed, missing and filled permanent teeth (DMFT) were calculated.

Consent was obtained from each patient before the commencement of treatment with ethical approval obtained through the University of Otago Ethics Committee (HD15/053).

Clinical outcome measures

The Peer Assessment Rating (PAR) index was used on models to assess improvement in the occlusion after orthodontic treatment.\(^2\)\(^2\) The index measures 11 occlusal traits, with each component score having a different applied weighting to calculate the overall score for each patient. The difference between the total pre- and post-treatment scores shows the percentage improvement, which can be classified as: changes <30% considered ‘worse/no different’, changes >30% ‘improved’, and changes > 21 PAR points are considered ‘greatly improved’. An accredited examiner scored all pre- and post-treatment orthodontic models using the standardised PAR ruler. Repeat measurements were taken for 10 cases one week following the initial assessment, and the intra-class correlation was 0.96, showing excellent intra-observer reliability. The orthodontic study models were available for 57 participants (95%) but three models were either lost or damaged.

Oral health-related quality of life (OHRQoL)

The short-form 8-item CPQ\(^{11-14}\) (Impact Short Form:8-ISF:8) questionnaire was completed before (T\(_0\)) and after (T\(_1\)) orthodontic treatment. The questionnaire is divided into four domains of oral symptoms, functional limitations, emotional and social well-being, with each domain having two items derived from the original 37-item CPQ.\(^3\) Patients were asked to recall the frequency of specific events during the past three months using a 5 point Likert scale for each item with ‘Never’ = 0; ‘Once/twice’ = 1; ‘Sometimes’ = 2; ‘Often’ = 3; and ‘Every day/almost every day’ = 4. A score was generated for each domain by totalling the individual item scores, to provide a final score ranging from 0–32. The higher the score the greater the negative impact of the oral conditions on quality of life.\(^6\)

For analysis purposes, the oral symptoms and functional limitations were combined into a single domain of ‘symptoms’ while emotional and social well-being were combined into a single domain of ‘wellbeing’.\(^2\)\(^3\) The questionnaires included global questions that asked the respondent to rate their oral health (health of their teeth, lips, jaws and mouth), with responses being ‘excellent’ (0), ‘very good’ (1), ‘good’ (2), ‘fair’ (4) and ‘poor’ (5), and their overall wellbeing (how much effect on their life overall), with responses being ‘not at all’ (0), ‘a little bit’ (1), ‘some’ (2), ‘a lot’ (3) or ‘very much’ (4).\(^2\)\(^4\)

Statistical analysis

Data were analysed using the Statistical Packages for the Social Sciences (SPSS version 20.0; IBM, NY, USA). Differences in proportions were tested for statistical significance using the Chi-square test, while differences in continuous variables were examined using the Kruskal-Wallis and Man-Whitney U tests. Effect sizes were used to determine the magnitude of the statistical difference in scores. These were calculated by dividing the mean change in pretreatment and post-treatment scores by the pooled standard deviations after correcting for the amount of correlation between the two scores.

Results

Table I shows the sociodemographic and malocclusion demographics by study group. Significant differences were found for mean baseline age, with the CL/P group being younger, and socioeconomic status, with the non-cleft group having a greater proportion in the high deprivation group. There were no differences in severity of the pretreatment malocclusion (DAI), dental health status (DMFT) or treatment duration.

The CPQ-ISF8 demonstrated a consistent gradient between the mean scores and the global question responses, indicating good construct validity (Table II). The mean T\(_0\), T\(_1\) changes in the overall and domain scores showed a consistent gradient of improvement (decrease in score) in both study groups, with the CL/P group having the largest mean difference (Table III).
Table I. Sociodemographic characteristics by study group (brackets contain percentages unless otherwise stated).

<table>
<thead>
<tr>
<th></th>
<th>Standard (N = 27)</th>
<th>CL/P (N = 33)</th>
<th>Combined (N = 60)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Sex</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>12 (44.3)</td>
<td>21 (63.6)</td>
<td>33 (55.0)</td>
</tr>
<tr>
<td>Female</td>
<td>15 (55.6)</td>
<td>12 (36.4)</td>
<td>27 (45.1)</td>
</tr>
<tr>
<td>Mean baseline age (years)</td>
<td>14.7 ± 1.5</td>
<td>12.1 ± 2.0 *</td>
<td>13.2 ± 2.2</td>
</tr>
<tr>
<td><strong>Ethnicity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>NZ European</td>
<td>18 (72.0)</td>
<td>23 (71.9)</td>
<td>41 (71.9)</td>
</tr>
<tr>
<td>Maori</td>
<td>3 (12.0)</td>
<td>4 (12.5)</td>
<td>7 (12.3)</td>
</tr>
<tr>
<td>Asian</td>
<td>4 (16.0)</td>
<td>2 (6.3)</td>
<td>6 (10.5)</td>
</tr>
<tr>
<td>Other</td>
<td>0 (0.0)</td>
<td>3 (9.4)</td>
<td>3 (5.3)</td>
</tr>
<tr>
<td><strong>Area-based SES</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low deprivation</td>
<td>11 (40.7)</td>
<td>14 (42.4)</td>
<td>25 (41.7)</td>
</tr>
<tr>
<td>Medium deprivation</td>
<td>5 (18.5)</td>
<td>14 (42.4)</td>
<td>19 (31.7)</td>
</tr>
<tr>
<td>High deprivation</td>
<td>11 (40.7)</td>
<td>5 (15.2) *</td>
<td>16 (26.7)</td>
</tr>
<tr>
<td><strong>Treatment need</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No need/ minor</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>Definite</td>
<td>0 (0.0)</td>
<td>4 (12.1)</td>
<td>4 (6.7)</td>
</tr>
<tr>
<td>Severe</td>
<td>0 (0.0)</td>
<td>4 (12.1)</td>
<td>4 (6.7)</td>
</tr>
<tr>
<td>Handicapping</td>
<td>27 (100)</td>
<td>25 (75.8)</td>
<td>52 (86.7)</td>
</tr>
<tr>
<td>Mean DAI Score</td>
<td>46 (6.2)</td>
<td>48 (15.1)</td>
<td>47 (11.9)</td>
</tr>
<tr>
<td>Mean DMFT</td>
<td>1.1 (1.7)</td>
<td>0.8 (1.2)</td>
<td>1.0 (1.4)</td>
</tr>
<tr>
<td>Mean treatment duration (months)</td>
<td>21.8 (9.5)</td>
<td>18.6 (9.6)</td>
<td>20.3 (9.6)</td>
</tr>
</tbody>
</table>

* p < 0.005
* three cases with missing data

Table II. Mean CPQ-ISF 8 score by global oral health question.

<table>
<thead>
<tr>
<th>Question</th>
<th>Mean CPQ-ISF8 score</th>
<th>SD</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>‘Would you say the health of your teeth, lips, jaws and mouth is’</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Excellent/ Very good</td>
<td>5.0</td>
<td>4.0</td>
<td></td>
</tr>
<tr>
<td>Good</td>
<td>9.3</td>
<td>4.7</td>
<td></td>
</tr>
<tr>
<td>Fair/Poor</td>
<td>10.6</td>
<td>4.3</td>
<td>0.011</td>
</tr>
<tr>
<td>‘How much does the condition of your teeth, lips, jaw or mouth affect your life overall?’</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Not at all</td>
<td>5.3</td>
<td>3.5</td>
<td></td>
</tr>
<tr>
<td>A little bit</td>
<td>8.0</td>
<td>3.6</td>
<td></td>
</tr>
<tr>
<td>Some/A lot/ Very Much</td>
<td>12.9</td>
<td>5.0</td>
<td>0.001</td>
</tr>
</tbody>
</table>

* 11 cases with missing data
A $p$-value $< 0.05$ was noted between $T_0$ and $T_1$ for item 2 (bad breath) for the CL/P group and 4 (difficulty with hot/cold) for the non-cleft group. There were no significant associations between CPQ score and dental health status (correlation coefficient = 0.118, $p = 0.381$), and no significant differences in CPQ change across the three SES deprivation categories of NZDep2013 ($p = 0.567$).

The changes in PAR component and weighted total scores following orthodontic treatment are shown in Table IV. There was a statistically significant difference in the change in PAR score and the proportion of individual components in each treatment outcome category. All of the non-cleft group had ‘improved’ (>30%) outcomes with the mean 29.5 PAR score change (‘greatly improved’ $>21$ points). In contrast, 22% of the CL/P group had ‘worse or no change’ (<30%) outcomes with a mean 16.1 PAR score change.

There was no significant correlation between changes in the PAR and CPQ-ISF8 (Pearson Correlation Coefficient -0.1, $p = 0.47$).

### Discussion

Several studies have investigated the OHRQoL for individuals with CL/P following orthodontic treatment in New Zealand, however the present study is the first to directly compare OHRQoL and orthodontic treatment outcome measures. It is noteworthy that the two study groups, which had comparable malocclusion severity, had similar OHRQoL changes following orthodontic treatment despite the fact the non-cleft group had better clinical treatment outcomes. The lack of correlation between OHRQoL and orthodontic status measures is consistent with other studies.25,26

The two global ratings of oral health and overall well-being demonstrated that CPQ-ISF8 had good construct validity, proving it to be a valid measure of OHRQoL for both patient groups. The baseline domain and total scores were worse for the non-cleft group, which was similar to the results of a previous study that used the OHIP14. The mean age difference between the two groups, with the non-cleft group 2.5 years older, may have influenced this
finding, although age does not appear to influence psychological well-being of CL/P patients. The non-cleft group had handicapping malocclusions as defined by the DAI, compared with 75.8% of the CL/P group, while the remaining patients were split between ‘severe’ and ‘definite’ malocclusions. In addition, there may also be an element of resilience and adaptation by the CL/P group, who were possibly more accepting of their oral condition than their non-cleft peers.

The two CPQ-ISF8 specific items that were statistically different ($p < 0.05$) at $T_0$ between the two groups were ‘bad breath’ and ‘difficulty with hot/cold’, with the non-cleft group presenting worse (higher) scores for both items. This is difficult to explain as there was no difference in dental health status between the two groups, as measured by the DMFT index, and respondents were only asked to report the frequency of oral symptoms and not severity. A previous study using the same questionnaire also found a large proportion of children without caries reported higher scores for ‘difficulty in eating with hot/cold foods and drinks’. It was also surprising to find only a relatively small improvement in the CPQ mean total and domain scores at $T_1$, with the CL/P group showing greater improvements. This contradicts the findings of a similar study in which significant improvements in OHRQoL were identified in non-cleft and orthognathic patients, and much smaller, non-significant differences were noted for CL/P, although the OHRQoL measure used was not child specific.

There were marked differences in the PAR Index $T_1$ scores between the two groups, with the CL/P group having considerably more variability and a worse mean score. This difference may reflect the difficulties in fully correcting the malocclusion given factors such as adverse growth following surgical repair leading to more prevalence of posterior crossbite and greater difficulty achieving Class I molar relationship on the cleft affected side. In addition, multiple dental anomalies often occur in the CL/P group making anterior correction difficult. This finding also raises the question of the most appropriate occlusal index for specific application to CL/P patients, but, to date, there is no other suitable alternative available. The percentage of the CL/P who experienced ‘worse or no different’ PAR percentage changes (22.6%) was disappointing and was greater than that reported for unilateral cleft lip and palate patients treated in the United Kingdom.

There are several factors that must be considered when interpreting the findings of the present study. The survey sample consisted of hospital-treated patients, who had more severe malocclusions than the average adolescent seeking orthodontic treatment in New Zealand, which therefore limited the general interpretation of the results. The eligibility criteria at the hospital also influenced the demographic characteristics of the study groups as the CL/P group were significantly younger, reflecting the priority of access to treatment for these patients and hence a shorter wait list time for this group. In addition, the difference in SES reflects the eligibility for orthodontic treatment of children without craniofacial anomalies.

It is known that the $T_0$ CPQ-ISF8 score is worse in patients with handicapping malocclusions and therefore, in the present study, their use as a comparison group may have masked the negative impact of CL/P on OHRQoL at $T_0$. It should also be noted that the relatively small sample of 60 patients may not have been adequately powered to detect differences between the groups and there were insufficient numbers in the CL/P group analysis the impact of different subtypes of cleft, hence the comparison of the results should be considered with caution.

Although the CPQ questionnaire is specific to children, it is not cleft specific and therefore may not address all aspects of the CL/P patients. There are a number of aspects of the CL/P condition that are not specifically addressed, including nasal deformity, speech problems and soft tissue scarring. The development of a self-reported outcomes measure for the cleft lip and palate population is inherently complicated due to a wide range of issues associated with this condition, as well as the potential range of ages of these patients under care. Since the present study commenced, more recent cleft specific questionnaires have been used with further developments in OHRQoL measures proposed. However, the use of the generic questionnaires, like the CPQ applied in the present study, allows for intergroup comparisons and benchmarking with other similar studies that examined heterogenic groups.

Future studies could consider using both generic and cleft specific assessment tools to obtain greater insight into the OHRQoL and clinical treatment outcomes, which may highlight specific factors influencing treatment outcomes. In addition, a long-term follow-up of these patients would also be beneficial in assessing the stability of OHRQoL and treatment changes.
Conclusion
The presence of CL/P highlighted no significant differences in baseline or changes in OHRQoL following orthodontic treatment compared with a non-cleft group. However, there were significant differences noted in clinical treatment outcome despite both groups having similar malocclusion severity. No significant correlation was noted between OHRQoL and clinical treatment outcome.

Acknowledgments
This study was undertaken with financial assistance from the New Zealand Dental Association Canterbury Branch Summer Studentship.

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