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Oral health-related quality of life in non-syndromic cleft lip and/or palate patients: a systematic review

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The Objective: To evaluate oral health-related quality of life (OHRQoL) in non-syndromic patients with cleft lip and/or palate (CLP), in comparison to a general non-cleft population. ***Basic Research Design:*** Systematic review. A literature search was conducted to identify papers reporting on OHRQoL in cleft samples. Only studies with suitable control groups were included. From each included paper were extracted the study and sample characteristics and results. ***Main outcome measures:*** OHRQoL score. ***Results:*** Three papers were chosen according to the preset inclusion and exclusion criteria. All used an OHRQoL generic patient-reported questionnaire with evidence of a development and validation process, with responses recorded on a five-point scale. The results could not be combined for the purposes of meta-analysis due to lack of standardisation. In 2 of the 3 studies, the OHRQoL was found to be significantly lower in the cleft than in the non-cleft samples (in patients 8-18 or 18-65 years of age). The third study, based on a relatively small sample size, could not detect significant differences between cleft and non-cleft individuals. ***Conclusions:*** Based on the results of the few studies included in the present systematic review, non-syndromic patients with CLP tend to have a lower OHRQoL than a general non-cleft population. This seems to hold true both for children and adults.

Key words*: cleft lip; cleft palate; quality of life; questionnaire; self report; review, systematic*

Introduction

According to a recent Centers for Disease Control and Prevention (CDC) report, cleft lip and/or palate (CLP) is the second most common birth defect, occurring in 1 in 575 live births (CDC, 2006). Children born with CLP may be affected by a combination of various facial differences, disturbances of the dentition and growth of the jaws, as well as swallowing, speech, and hearing disorders (Klassen *et al*., 2012). The treatment of these differences usually involves numerous complex and lengthy procedures from infancy into adulthood, but the ultimate goal is to achieve a good aesthetic and functional result which will allow for psychological and social well-being.

The World Health Organization (WHO) defines health as a complete state of physical, mental, and social well-being and not simply the absence of disease (WHO, 1948). The concept of quality of life (QoL) is used to evaluate general well-being, and includes all emotional, social, and physical aspects of an individual's life. Within the field of medicine and healthcare, the term health-related quality of life (HRQoL) is used, referring to how the individual's well-being may be impacted over time by a disease, disability, or disorder. When oral health conditions impact on an individual's well-being, the evaluation of the particular oral health condition in question as it interacts with the individual's well-being is referred to as oral health-related quality of life (OHRQoL).

OHRQoL is a multidimensional construct that includes a subjective evaluation of the individual's oral health, functional well-being, emotional well-being, expectations and satisfaction with care, and sense of self (Sischo and Broder, 2011). OHRQoL is an integral part of general health and well-being. It is recognised by the WHO as an important segment of the Global Oral Health Program (WHO, 2003). Since Cohen and Jago (1976) first advocated the development of sociodental indicators, a variety of efforts have been invested in developing instruments to measure OHRQoL.

The Surgeon General in the USA has identified OHRQoL as a health priority (DHHS, 2000) and QoL is now at the forefront on public health policy (Sischo and Broder, 2011). Including OHRQoL in research adds a powerful dimension in the planning and development of health promotion programmes and by identifying groups who are vulnerable for low OHRQoL, investigators can use data from this research to create programmes aimed at improving oral health and elevating OHRQoL (Sischo and Broder, 2011). With increasing focus on health policy to address health promotion and disease prevention, OHRQoL has come to incorporate both positive and negative perceptions of oral health and health outcomes (Broder and Wilson-Genderson, 2007).

A workshop entitled "Prioritizing a research agenda for orofacial clefts" was conducted in 2006 at the CDC by the National Center on Birth Defects and Developmental Disabilities to review the knowledge on orofacial clefts and identify the knowledge gaps that need additional public health research (Yazdy *et al*., 2007). They created a prioritised public health research agenda based on these gaps, which could be of value in guiding future research in the area of orofacial clefts. One of the knowledge gaps identified was QoL for children with orofacial clefts (Yazdy *et al*., 2007).

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Complimentary to this recent effort is a review of the impact of orofacial clefts on QoL and healthcare use and costs (Wehby and Cassell, 2010), which aimed to identify primary research gaps in this field and potential study designs to address these gaps. The authors here also recommended that a need exists for evaluating the impact of clefts on HRQoL of affected individuals and families throughout the lifespan, using large population-based samples, robust measures and multiple perspectives including the societal perspective.

In the absence of CLP specific validated questionnaires, many studies employ generic, non-cleft-specific, questionnaires which measure OHRQoL. These instruments, although reliable, are less likely to be sensitive to all the issues specific to the CLP population and the changes resulting from treatment (Eckstein *et al*., 2011). The needs of individuals with clefts are unique and instruments to measure the OHRQoL specific to individuals with clefts are needed. However, this limits the comparability of OHRQoL assessments across disorders and to controls. Despite the fact that measuring OHRQoL can be difficult, universal instruments and data collection from across centres for cleft treatment could make the task easier. In addition a better understanding of how individual components such as speech and facial appearance correspond to overall measures could help identify ways to improve OHRQoL for individuals with clefts (Yazdy *et al*., 2007).

Questionnaires completed by the patient in question aim to quantify QoL and other significant outcome variables (such as satisfaction, symptoms, and function) from the patient's perspective (Pusic *et al*., 2011). To encompass all questionnaires completed by patients, the umbrella term patient-based outcome measures (PBOs) has been suggested by Fitzpatrick *et al*. (1998) on the grounds that all are dependent upon what patients have to say about their health.

On the subject of QoL in patients with CLP, two recent systematic reviews have been carried out, one looking into the psychosocial effects of CLP (Hunt *et al*., 2005) and the other looking into the QoL of children treated for CLP (Klassen *et al*., 2012). However, no systematic review has been carried out to date looking specifically into OHRQoL in patients with CLP. The purpose of the present investigation was to systematically review and evaluate OHRQoL in non-syndromic patients with CLP, in comparison to a general non-cleft population.

Methods

Protocol and registration

When planning and carrying out the present systematic review, the preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines were followed as closely as possible (Liberati *et al*., 2009). Methods of analysis, inclusion and exclusion criteria, and the main outcome measure were defined in advance of the study. However, a review protocol was not published nor was the study registered.

Eligibility criteria

Studies evaluating OHRQoL in non-syndromic CLP patients were investigated. Trials were retrieved with no date, language, or publication status restriction. The method under evaluation was the use of PBOs, namely questionnaires, to quantitatively evaluate OHRQoL.

Inclusion criteria were: 1, cross-sectional or longitudinal studies evaluating OHRQoL in CLP patients; 2, the presence of a suitable control group (healthy non-cleft individuals); 3, the use of an OHRQoL PBO, namely a questionnaire; 4, the presentation of results for cleft patients and non-cleft patients presented separately.

Exclusion criteria were: 1, case reports or case series (sample size ≤10); 2, papers without a suitable control group: 3, studies involving syndromic cleft patients; 4, studies involving other craniofacial anomalies or dentofacial deformities; 5, the use of interviews (structured, semi-structured, or unstructured), and not questionnaires; 6, the use of outcome measures where the parent, caregiver, health professional, or any other person besides the patient reported the outcome; 7, the use of an *ad hoc* patient-reported outcome questionnaire (i.e., one without published evidence of a development or validation process). In light of the fact that investigations looking into OHRQoL in CLP patients were not numerous, studies were not excluded on the basis of poor methodological quality.

The main outcome was the OHRQoL score in CLP patients at any given time point (no limitation regarding the age of the subjects was imposed), in comparison to a non-cleft control group.

Information sources and search

Relevant studies were located by searching the following databases: Pubmed, EMBASE, Scopus, Web of Science, CINAHL and the Cochrane Library. The 'related citations' function in Pubmed was used to retrieve further papers, as was citation tracking. Authors' names that appeared on numerous occasions in the literature search were additionally searched to retrieve any further papers. The reference lists of the retrieved papers were hand searched to identify studies that might not have been included. The last search was conducted in August 2012.

The search and study selection was carried out independently by two reviewers. The terms used in the search strategy were the following: 1, quality of life, QoL, health-related quality of life, HRQoL, oral health-related quality of life, OHRQoL; 2, cleft, craniofacial, orofacial, dentofacial, lip, palate. Searches were conducted using combinations of one of the terms from the first search category with one of the terms from the second search category. Searches were also conducted using combinations of one of the terms from the first search category with specific measures of OHRQoL (the full name or the abbreviation) including:

- activities of daily living;
- child oral health impact profile;
- child oral health quality of life questionnaire;
- child oral impacts on daily performances;
- child perception questionnaire;
- dental impact on daily living;
- dental impact profile;
- general oral health assessment index;
- geriatric oral health assessment index;
- oral health impact profile;
- oral impact on daily living;
- oral impacts on daily performances;
- OH-QoL UK;
- social impacts of dental disease;
- sociodental scale;
- subjective oral health status indicators;
- surgical orthodontic outcome questionnaire.

Study selection

Titles and abstracts of the papers were initially evaluated. If eligibility could not be determined based on this, full texts of the papers were retrieved. Full-text papers were assessed for eligibility by applying the inclusion and exclusion criteria. Finally, eligible studies were collected for data extraction. If the two reviewers could not agree on the eligibility of a certain study, disagreements were resolved by discussion.

Data collection process and data items

From each included study the following information was extracted: publication data, study design, sample characteristics, control group characteristics, treatment history and characteristics, outcome measure (PBO) used, and OHRQoL data. Disagreements were resolved by discussion between the reviewers.

Risk of bias within individual studies was assessed by considering five different domains of bias defined by the Cochrane Bias Methods Group: namely selection; performance; detection; attrition; and reporting. Additionally, an assessment of confounding factors was carried out. The judgement for each of the domains was formulated by answering a pre-specified question, such that an answer of yes indicated low risk of bias, an answer of no indicated high risk of bias, and an answer of unclear indicated unclear or unknown risk of bias. The pre-specified questions were as follows:

- Selection: Did the chosen sample include all of the possible cases or a random selection of representative cases?
- Performance: Were the experimental sample and control groups exposed only to the intervention of interest and not other factors?
- Detection: Were the self-reported questionnaires completed by the subject in a private environment without outside influence?
- Attrition: Was there description of the completion of outcome data including attrition and exclusions from the analysis?
- Reporting: Was reporting complete and not selective, with presentation of the results for the different domains of the questionnaire?
- Confounding: Were confounding factors assessed such as ethnicity, socioeconomic status, other oral health issues, and other health issues?

Following recommendations from the Cochrane Collaboration, the methodological quality of included studies was assessed using the Newcastle-Ottawa Scale (Wells *et al*., 2013). This instrument assesses the quality of non-randomised studies in three broad study design categories: namely patient selection, comparability of study groups, and assessment of outcome. A star system is used where each study can be awarded a maximum of 8 stars. For the purposes of this systematic review, studies with a score of 0-2 were considered low quality, 3-5 medium quality, and 6-8 high quality.

In addition to the quality assessment, an evaluation of the included studies was carried out to determine the degree to which they met the minimal standards for reporting for cross-sectional studies using PBOs, as defined by Tsakos *et al.* (2012).

Summary measures and synthesis of results

The difference in means was the intended main summary measure, comparing the cleft sample to the non-cleft control sample. We intended to combine the results using the random-effects model of meta-analysis, and arrive at standardised mean differences and 95% confidence intervals of the main outcome. We also intended to carry out heterogeneity tests, using I^2 and evaluate publication bias by visual inspection of the funnel plots.

Results

Study selection

The initial database search yielded 1033 papers. After preliminary exclusion, 97 papers remained and were screened for eligibility. Following exclusion on the basis of the content of the abstract, 23 papers were evaluated in their full-text form. Strict application of the inclusion and exclusion criteria provided 3 studies (Foo *et al*., 2012; Ward *et al*., 2012; Wogelius *et al*., 2009) for inclusion in this systematic review (Figure 1).

Study characteristics

The characteristics of the included studies are presented in Table 1. All of the studies were cross-sectional in nature, and used an OHRQoL generic patient-reported outcome measuring instrument (namely a questionnaire) with published evidence of a development and validation process, with responses recorded on a five-point scale. The included studies involved a total of 199 cleft lip and/or palate patients and 4496 control patients. The range of ages of the combined samples from the individual studies was from 8 to 65 years, both for the cleft and for the non-cleft samples.

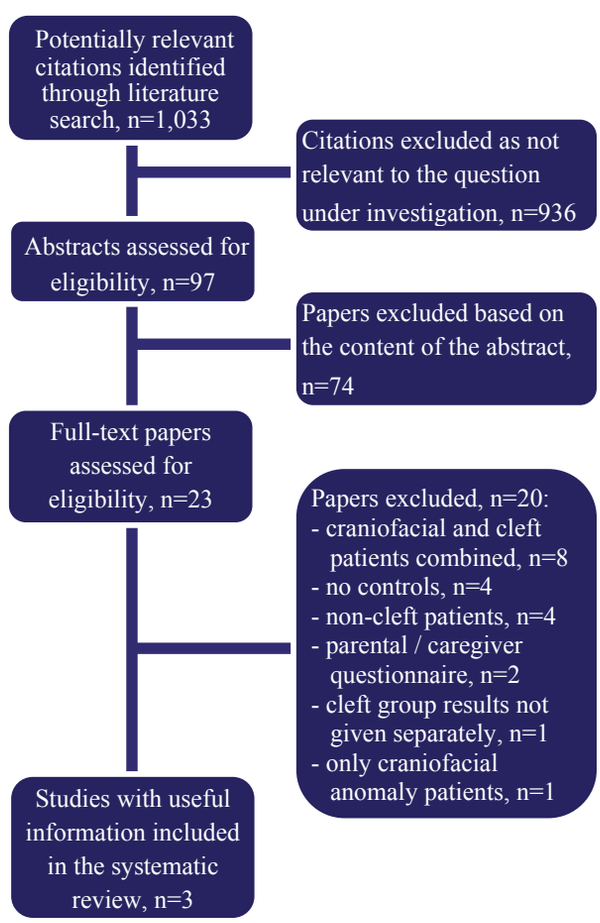

Figure 1. *Flow diagram summarising the literature search*

Table 1. Characteristics of the included studies

Authors, year of publication	*Origin of population*	*Study n*	*population, age (years)*	*Cleft distribution*	*Surgical procedures undertaken*	*Control n*	*population, age (years)*	*Type of control*	*OHRQoL measure*
Foo *et al*., 2012	Australia	88 40 female; 48 male	range 18-65 ≤27 (n=44); ≥28 (n=44) mean 31; median 27.5 range 8-18	52 UCLP; 32 BCLP 2 CP; 1 SC	for cleft for jaw discrepancies	4170	age-matched against study population	representative Australian population (Slade *et al*., 2007)	OHIP-14
Ward *et al*., 2013	USA	75 27 female; 48 male	8-10 (n=22) 11-14 (n=35) 15-18 (n=18)	25 UCLP; 21 BCLP 14 UCL; 3 BCL 7 CP; 5 CSP	NS	75 46 female; 29 male	range 8-18 8-10 (n=20) 11-14 (n=15) 15-18 (n=40)	patients from: - pediatric primary care clinic - adolescent medicine clinic	COHIP
Wogelius *et al*., 2009	Denmark	15 21	range 8-10 range 11-14	NS NS	NS NS	97 154	range 8-10 range 11-14	healthy public school children	CPQ8-10 CPQ11-14

Abbreviations: BCL, bilateral cleft lip; BCLP, bilateral cleft lip and palate; COHIP, 38-item Child Oral Health Impact Profile questionnaire; CP, cleft palate; CPQ8-10 , Child Perceptions Questionnaire for children 8-10 years; CPQ11-14 , Child Perceptions Questionnaire for children 11-14 years; CSP, cleft soft palate; n, sample size; NS, not specified; OHIP-14, Short-form (14-item) Oral Health Impact Profile questionnaire; OHRQoL, oral health-related quality of life; SC, submucous cleft; UCL, unilateral cleft lip; UCLP, unilateral cleft lip and palate

The reporting of information in individual studies was not always complete. For example, not all studies reported on the distribution of cleft type, the gender distribution, or the mean age for each group.

The quality assessment revealed that, based on the criteria for cross-sectional studies, one study was judged to be of high quality (Foo *et al*., 2012) while the other two were judged to be of moderate quality (Ward *et al*., 2012; Wogelius *et al*., 2009). The evaluation of the minimal standards for reporting for cross-sectional studies using PBOs is presented in Table 2. None of the studies met all of the requirements.

Results of individual studies, bias, and synthesis of results

An assessment of within study bias is presented in Table 3. A certain amount of bias was present in all included studies, with none of the studies formally accounting for confounding factors such as ethnicity, socioeconomic status, or the presence of other general health or oral health issues.

The results from the studies could not be combined for the purpose of a meta-analysis due to the lack of standardisation among studies. Thus, neither heterogeneity tests nor an

assessment of between study bias were carried out because meta-analysis methodology was not used to combine results.

The summary of the results from each individual study as regards OHRQoL are presented in Table 4. Results for both the cleft patients as well as the control samples are shown. Since a combination of results was not possible, the main results from each study relevant to the current investigation are summarised.

Foo *et al*. (2012), using the Short-form (14-item) Oral Health Impact Profile questionnaire (OHIP-14: Slade and Spencer, 1994; Slade, 1997) on 18-65 year old individuals, found that OHRQoL was significantly worse for cleft patients than for the general population, observed by a higher OHIP-14 mean score in cleft patients (lower scores reflect a better OHRQoL). The score was 1.7 times higher for cleft patients than the general population estimates. No significant differences were found by age or by gender. They conclude that treatment for orofacial clefting does not entirely remove factors contributing to poor OHRQoL.

Ward *et al*. (2012), using the 38-item Child Oral Health Impact Profile questionnaire (COHIP: Broder *et al*., 2007; Broder and Wilson-Genderson, 2007) on 8-18 year olds, found that OHRQoL was significantly worse for cleft patients than for non-cleft patients, determined by a lower COHIP score in cleft patients (higher scores reflect a more positive OHRQoL). The difference in the overall COHIP scores was due to significant differences in the functional well-being and social-emotional well-being domains of the questionnaire.

Wogelius *et al*. (2009), using the Child Perceptions Questionnaire for 8-10 years ($CPQ_{8\text{-}10}$: Jokovic *et al*., 2004) and the Child Perceptions Questionnaire for 11-14 years ($CPQ_{11\text{-}14}$: Jokovic *et al*., 2002; 2006; Locker *et al*., 2005), found no significant differences between cleft patients and healthy school children of the same age.

Table 2. Minimum reporting standards for cross-sectional studies using patient-based outcome measures

Minimum reporting standards	*Foo et al., 2012*	*Ward et al., 2013*	*Wogelius et al., 2009*
Description			
Mean/Median	✓	✓	✓
Alternative scoring formats	✓	--	✓
Interpretation			
Statistical significance	✓	✓	✓
Effect size	✓	--	--
Standard error of measurement	--	--	--
Global ratings (oral health/quality of life)	--	✓	✓
Well-established clinical groups/benchmarks	✓	✓	✓

Table 3. Risk of bias of various types within individual studies

Types of bias	*Foo et al., 2012*	*Ward et al., 2013*	*Wogelius et al., 2009*
Selection bias	Low risk	Low risk	High risk
Performance bias	Low risk	High risk	Low risk
Detection bias	Low risk	Unknown	Unknown
Attrition bias	Low risk	Low risk	High risk
Reporting bias	High risk	Low risk	Low risk
Confounding bias	High risk	High risk	High risk

Table 4. OHRQoL scores from individual studies

Studies' authors, year of publication	*Ages of patients (years)*	*Questionnaire used*	*Cleft sample (n)*	*Overall respective OHRQoL score*				
				Cleft sample		*Control sample*		*Significance*
				mean	*variation*	*mean*	*variation*	
Foo *et al*., 2012	18-65	OHIP-14	88	12.7	10.7-14.7 (95% CI)	7.5	7.2-7.9 (95% CI)	yes (non-overlapping CIs)
Ward *et al*., 2013	8-18	COHIP	75	95.6	sd 18.3	108.6	sd 16.9	yes (p=0.01)
Wogelius *et al*., 2009	8-10	CPQ8-10	15	7.9	sd 8.0	8.5	sd 6.2	no (p=0.50)
	11-14	CPQ11-14	21	10.2	sd 7.2	10.5	sd 7.6	no (p=0.84)

Abbreviations: CI, confidence interval; COHIP, 38-item Child Oral Health Impact Profile questionnaire; CPQ8-10 , Child Perceptions Questionnaire for children 8-10 years; CPQ11-14 , Child Perceptions Questionnaire for children 11-14 years; OHIP-14, Short-form (14-item) Oral Health Impact Profile questionnaire; sd, standard deviation

Discussion

The results of the present systematic review suggest that non-syndromic CLP patients tend to have a lower OHRQoL than a general non-cleft population. This seems to hold true, on the whole, both for children (8-18 years of age) as well as for adult individuals (18-65 years of age). Very few studies were included however in the present systematic review, which highlights the shortage of high-quality data in this field.

Methodological quality of the included studies was moderate on average, with only one of the studies judged as being of high quality. The risk of bias however was present in all of the included studies to a certain extent, which must be taken into account when interpreting results. One of the studies included in the present systematic review was based on a relatively small sample size concluding that no significant differences could be detected between cleft and non-cleft children (in patients 8-10 or 11-14 years of age). On the other hand the other two studies with their larger sample sizes, noted worse OHRQoL in cleft compared to non-cleft individuals. Study findings are sometimes inconsistent, and it has been suggested that this is partly because of differences between studies with regard to patient populations, HRQoL measures used, and study design (Wehby and Cassell, 2010). In the literature to date, the most common way of presenting data from OHRQoL studies is in terms of aggregate arithmetic scores, along with tests of statistical significance of differences between groups or between time points. The use of a single aggregate score is not without limitations however. A given score can be derived from different sets of responses with different items affected to a varying degree, therefore making it impossible to provide one profile for a specific score (Tsakos *et al*., 2012). This emphasises the need to present OHRQoL data also using alternative scoring formats. One of the studies included in the present systematic review (Foo *et al*., 2012) used three summary variables which can assist in the communication of results. The three variables were computed as follows: 1, prevalence: the percentage of people reporting one or more items "fairly often" or "very often"; 2, extent: the number of items reported "fairly often" or "very often"; 3, severity: the sum of ordinal responses. The use of these or similar alternative OHRQoL scores could be employed more often, improving the appraisal and appreciation of presented data.

A recent study (Eckstein *et al*., 2011), aiming to identify and assess the extent to which currently existing PBOs, validated in a CLP population, met internationally established criteria based on guidelines set by the Scientific Advisory Committee of the Medical Outcomes Trust for health-related outcome measures (Scientific Advisory Committee of the Medical Outcomes Trust, 2002), found that only two OHRQoL measures (namely the COHIP and CPQ) were sufficiently validated according to guidelines. This finding may impel authors to use these measures to carry out OHRQoL studies in the future.

In any study exploring OHRQoL, it is important to identify whether a statistically significant difference between experimental and comparison groups has clinical meaningfulness, and whether a statistically significant difference at the group level has relevance for clinically meaningful change at the individual level (Sischo and Broder, 2011; Tsakos *et al*., 2012). This is often difficult to define and assess at an individual and a study level, making interpretability of OHRQoL data difficult (Tsakos *et al*., 2012). More well-designed controlled studies need to be carried out using scientifically sound and validated questionnaires, with better quality reporting, which will allow more data to be collated, and the minimally important difference (Jaeschke *et al*., 1989) to be established.

It has been highlighted that a tremendous need exists for expanding the collaborations between various birth defect registries, craniofacial care providers, and research to identify data needs, improve data collection systems, and build consortia that provide access opportunities to further examine the impact of clefts on multiple outcomes throughout the lifespan (Wehby and Cassell, 2010). One such consortium, the FaceBase Consortium (Hochheiser *et al*., 2011), although not geared towards QoL research but rather towards cleft and craniofacial research (e.g. anatomical, developmental, molecular, genetic) provides an example of how a consortium can be created and run. This could be used as a model of a comprehensive collaborative programme which could facilitate the development of a similar consortium to collect QoL data. To this end, the international community must agree on a strategy which facilitates comparison of data, and create national norms for frequently used measures (Allen, 2003). The array of existing measures must be compared and equivalency of scores determined, which will allow for recommendations and an evaluation of the appropriateness of their use in different circumstances and for different purposes (Weintraub, 1998).

Data from previous studies often lacks a comparison to a non-cleft control group, but findings from these studies can still provide valuable information. Bos and Prahl (2011) divided 8-15 year old CLP patients into cleft subgroups (cleft palate; cleft lip with or without cleft alveolus; unilateral cleft lip and palate; bilateral cleft lip and palate) and compared OHRQoL in each subgroup. They found that patients in the cleft lip and alveolus subgroup had higher scores for the functional well-being component of the COHIP, than the other subgroups. Munz *et al*. (2011) studied adolescent/young adult CLP patients having recently completed their last phase of surgical treatment and found that the more satisfied the patients were with their treatment and treatment outcomes, the more positive was their OHRQoL.

A recent six-centre NICDR-supported investigation (Broder *et al*., 2012) examining OHRQoL among school-aged children with cleft conditions, found that Black and mixed ethnicity youths with clefts had lower OHRQoL than did their White and Asian counterparts. Likewise, patients without private health insurance reported lower OHRQoL than did those with private health insurances. Finally, youths with surgical recommendations also had lower OHRQoL than did those without such surgical recommendations. Such findings suggest that vulnerable youths with clefts are at risk for reduced OHRQoL and unmet needs.

Despite the growing literature, as the current systematic review draws attention to, there is still a shortage of high-quality data on OHRQoL among individuals with orofacial clefting conditions. In an era of evidence-based care, this identifies the need to carry out well-designed and controlled studies in this field, and to present results and report outcomes in an adequate manner. Non-syndromic CLP patients need to be looked at in comparison to non-cleft control groups, and longitudinal research designs should be applied

to assess the effect of various treatments on OHRQoL and the changes in OHRQoL with time. This will further help establish cleft care standards. The division of CLP patients into cleft subgroups would also provide valuable information helping to identify which type of cleft is associated with better or worse OHRQoL outcomes.

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