

Orofacial functions and quality of life in children with unilateral cleft lip and palate

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Abstract: This study aimed to evaluate the orofacial functions and oral health-related quality of life (OHRQoL) of children with unilateral cleft lip and palate (UCLP). This case-control study included patients with UCLP matched by sex and age with controls (children without UCLP), resulting in the inclusion of a total of 108 eight- to ten-year-old children. Orofacial functions and OHRQoL were evaluated using the Nordic Orofacial Test-Screening (NOT-S) and the Child Perceptions Questionnaire (CPQ₈₋₁₀), respectively. Data normality was assessed by the Kolmogorov-Smirnov test. Differences and correlations in NOT-S and CPQ₈₋₁₀ scores between and within the groups were evaluated using Mann-Whitney and Spearman's correlation tests, respectively. The distribution of NOT-S and global ratings of CPQ₈₋₁₀ for each group were assessed by Chi-squared/Fisher's Exact tests. The UCLP group had a higher NOT-S total and examination scores than the controls. Dysfunctions related to breathing, facial symmetry/expression, and speech were more frequent in the UCLP patients than in the controls. The UCLP group had higher scores on the social well-being domain than the controls. There was a significant difference between the groups in their ratings in regards to the extent to which their oral condition affected their life overall, with controls perceiving it as somewhat better than patients. In both groups, NOT-S total and interview scores were positively correlated with CPQ₈₋₁₀ total and domain scores. The NOT-S examination score was only significantly correlated with social domain scores in the control group. The presence of UCLP was associated with clinical signs of orofacial dysfunctions related to breathing, facial symmetry/expression, and speech. Children with UCLP reported more orofacial dysfunctions and negative impacts on social well-being than controls.

Keywords: Cleft Lip; Cleft Palate; Disability Evaluation; Facial Asymmetry; Quality of Life.

Introduction

Cleft lip and palate (CLP) is the second most common birth defect and the most common congenital craniofacial abnormality, affecting, on average, one in 575 to almost 8 in 1,000 newborns.^{1,2,3} CLP can result in occlusal and anatomical alterations, most frequently a crossbite and skeletal

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class III malocclusion,⁴ and also seems to negatively impact the oral health-related quality of life (OHRQoL) of the affected children.⁵ Although demographic, socioeconomic, behavioral and psychosocial factors have been linked to OHRQoL, there is currently no agreement on the predictors in general and specific populations.⁶ Previous studies found a worse OHRQoL among Syrian⁷ and Brazilian children with CLP.⁸ Moreover, while some studies have suggested no differences for OHRQoL scores between children with or without CLP,⁹ others have reported lower negative impacts among CLP patients.¹⁰

Actually, the perceived OHRQoL measures may be considered as additional components to evaluate treatment of CLP patients in a more comprehensive way, considering that the assessment of outcomes frequently raises concerns about facial appearance, speech, and social impairments.¹¹ In a recent systematic review and meta-analysis, the presence of CLP negatively affected the health-related quality of life (HRQoL) of children and adults, mainly on the psychological health and vitality dimensions.⁶ On the other hand, no significant differences were found considering oral health and oral conditions among the children in the evaluated studies. These results could be explained by the wide range of age of the subjects, which may have blended the different perceptions of OHRQoL among childhood and adolescence.¹² Age-specific self-report measures are required to adapt differences in children's self-concepts, understanding of their feelings, and their ability to interpret other people's behavior. In terms of cognitive, emotional, social and language development, the age group of 8 to 10 years is fairly homogeneous.⁵ Despite this knowledge, there is a lack of studies about factors associated with the perception of OHRQoL related to CLP.

There are a number of distinct domains of OHRQoL measures involved in CLP children, such as appearance, speech, facial growth, and psychosocial interactions.¹³ In the Leme et al.¹⁴ study, the presence of orofacial dysfunctions was associated with worse OHRQoL in otherwise healthy children, suggesting that oral and craniofacial diseases or disabilities may contribute to the compromise of vital actions (e.g., breathing, chewing, and swallowing) and

muscle posture (e.g., mouth and tongue posture) and undermine the necessary requirements for social interactions, including speech, emotional communication, facial expression, and appearance. Notwithstanding, while there are existing studies evaluating the speech, muscle function, and facial asymmetry of children with CLP,^{15,16,17,18} there is no prior study correlating oral functions with impacts on OHRQoL. Considering the high prevalence of malocclusions among children with CLP, mainly a posterior crossbite that can result in facial asymmetry, abnormal tongue posture, an abnormal swallowing pattern, and temporomandibular disorders in adulthood,^{19,20} it is necessary to evaluate the orofacial functions of these patients as well as their correlations with OHRQoL.

Therefore, the present study aimed to evaluate the orofacial functions and OHRQoL of children with unilateral cleft lip and palate (UCLP). The research hypotheses were: a. orofacial dysfunctions affect children with UCLP and b. orofacial dysfunctions are correlated with OHRQoL.

Methodology

Ethical approval was obtained from the Human Research Ethics Committee of the Hospital for Rehabilitation of Craniofacial Anomalies (HRAC), University of São Paulo (USP), Bauru, SP, Brazil (protocol n. 318.871/2013). Participants and their parents were informed about the examination procedures and assured of the confidentiality of the collected information. Only those who signed the informed consent form were included in the study.

Study Design and Sample Characteristics

This case-control study was carried out among low-socioeconomic-status children aged 8 to 10 years. The children with UCLP (case group) were enrolled at HRAC/USP and the control group was represented by a population of children without CLP recruited from public schools of Piracicaba, SP, Brazil. The exclusion criteria for children with UCLP were tooth decay (n = 38), current use of an orthodontic appliance (n = 74), syndromes associated with CLP (e.g., Treacher Collins, Van der Woude, Pierre Robin, Apert and

others) ($n = 14$)²¹, and missing a consultation ($n = 41$). Exclusion criteria for the control group were tooth decay and current use of an orthodontic appliance. Thus, this study was conducted on 108 children matched by sex and age, resulting in 54 matched pairs. Data collection was conducted from July 2013 to November 2014.

Sample Size Calculation

The sample size was calculated on the basis of the NOT-S total score reported in a pilot study conducted with UCLP children enrolled in the HRAC/USP as described in our previous publication.²² Considering a mean of 0.24 NOTS total score, standard deviation of 0.9, a sampling error of 10%, and a confidence level of 95%, the required sample size was defined as 54 individuals in each group.

Orofacial Functions

Orofacial functions were evaluated in the children with UCLP and the controls using the Brazilian Portuguese version¹⁴ of the Nordic Orofacial Test-Screening (NOT-S) protocol.²³ This protocol consists of a structured interview and a clinical examination, each part including six domains. In the interview, the following functions were assessed: (I) sensory function, (II) breathing, (III) habits, (IV) chewing and swallowing, (V) drooling, and (IV) dryness of the mouth. In the examination, the following functions were assessed: (1) face at rest, (2) nose breathing, (3) facial expression, (4) masticatory muscle and jaw function, (5) oral motor function, and (6) speech. Each domain contains one to five items, reflecting the complexity of the specific function. The NOT-S was applied individually to each child by the same trained researcher (ABMM), in a vacant room, with the child seated in an upright position. The NOT-S interview was held by asking the questions printed on the screening form. To assess orofacial dysfunction in the clinical examination, the subjects were requested to carry out tasks for each item in conjunction with the illustrated manual. Each item has criteria for the respective function. An answer of YES or a task that met the criteria for impaired function resulted in a score of 1, indicating a dysfunction in the scored domain. An answer of NO or a task

that did not meet the criteria resulted in a score of 0 (zero). The total score was the sum of the score for each domain, ranging from 0 to 12. The higher the score, the worse the orofacial dysfunction.

Oral Health-Related Quality of Life

The impact of oral conditions on the OHRQoL of children with UCLP and controls was evaluated using the Brazilian Portuguese version²⁴ of the CPQ₈₋₁₀.⁵ It is a self-completed questionnaire with 25 items grouped into four domains: oral symptoms, functional limitations, emotional well-being, and social well-being. A Likert-type scale was used with the response options of "Never" = 0, "Once or twice" = 1, "Sometimes" = 2, "Often" = 3, and "Very often" = 4. The recall period was four weeks. The minimum possible score is zero and the maximum possible score is 100. A high score indicates more negative impacts on the OHRQoL.

Participants were also asked to give overall or global assessments of their oral health (OH) and the extent to which the oral or orofacial condition affected their overall well-being (OWB). These questions preceded the multi-item scales in the questionnaire. OH and OWB questions are worded as follows: "When you think about your teeth or mouth, would you say that they are..." and "How much do your teeth or mouth bother you in your everyday life?" These global ratings had a 4-point response scale ranging from "Very good" = 0 to "Poor" = 3 for OH, and from "Not at all" = 0 to "A lot" = 3 for OWB, respectively.

Statistical analysis

Statistical analysis was performed using SPSS 22.0 (SPSS Inc., Chicago, IL, USA) and BioEstat 5.0 (Mamirauá, Belém, PA, Brazil) with a 5% significance level. Data normality was assessed using the Kolmogorov-Smirnov test. Differences in NOT-S and CPQ₈₋₁₀ scores between the UCLP and control groups were evaluated by means of the Mann-Whitney test. The magnitude of the difference between groups was assessed using the effect size (ES). This was derived from the mean difference in scores between the groups divided by the pooled SD of scores: a value of 0.2 was taken to be small, 0.5 to be moderate, and 0.8 to be large.²⁵

The distribution of the NOT-S domain scores and responses to global ratings of CPQ₈₋₁₀ for each group was assessed by Chi-squared or Fisher's Exact tests. The correlation between NOT-S and CPQ₈₋₁₀ scores for each group was evaluated by Spearman's correlation test.

Results

This study evaluated orofacial functions and OHRQoL in 54 children with UCLP, matched by sex ($\chi = 57.41\%$) and age (9.07 ± 0.80 years-old) with controls. Table 1 shows the comparison of the NOT-S scores between the groups. The UCLP group had a higher mean NOT-S total score (4.3 vs. 2.8, $p < 0.0001$) and examination (2.2 vs. 0.5, $p < 0.0001$) score than the controls.

The distribution of the NOT-S domains for each group are presented in Table 2. Children with UCLP reported more dysfunction related to breathing than controls (29.6% vs. 7.4%, $p < 0.05$). All of these reports were related to snoring when sleeping. Clinical dysfunctions related to face at rest (100% vs. 13%; $p < 0.0001$), facial expression (63% vs. 22.2%, $p < 0.01$), and speech (77.8% vs. 27.8%, $p < 0.01$) were more frequent among children with UCLP than the controls. Asymmetry (100%) and deviant lip position (63%) were the most frequent dysfunctions clinically diagnosed in patients when the face was at rest. More than half of the patients (57.4%) could not pout or round their lips symmetrically (facial expression domain) and 74.1% presented with unclear speech in regards to one or more indistinct sounds or abnormal nasality.

Table 1. Comparison of the NOT-S scores between groups.

Variable	UCLP group	Control group	p-value*
	Mean (SD)	Mean (SD)	
Total scale [0–12]	4.3 (1.5)	2.2 (1.7)	< 0.0001
Interview [0–6]	1.6 (1.2)	1.7 (1.3)	0.474
Examination [0–6]	2.8 (0.9)	0.5 (0.7)	< 0.0001

NOT-S: nordic orofacial test-screening; UCLP: unilateral cleft lip and palate; sd: standard deviation. Values in square brackets indicate range of possible scores. *p-value obtained from the Mann-Whitney test.

Table 2. Distribution [n (%)] of the NOT-S domains by group.

NOT-S domains	UCLP group	Control group	p-value
Interview			
(I) Sensory function	4 (7.4)	5 (9.3)	1.000 ^b
(II) Breathing	16 (29.6)	4 (7.4)	0.025 ^a
(III) Habits	17 (31.5)	35 (64.8)	0.058 ^a
(IV) Chewing and swallowing	38 (70.4)	34 (63.0)	0.831 ^a
(V) Drooling	2 (3.7)	2 (3.7)	1.000 ^b
(VI) Dry mouth	7 (13.0)	12 (22.2)	0.418 ^a
Examination			
(1) Face at rest	54 (100.0)	7 (13.0)	< 0.0001 ^a
(2) Nose breathing	0 (0.0)	2 (3.7)	0.495 ^b
(3) Facial expression	34 (63.0)	12 (22.2)	0.010 ^a
(4) Masticatory muscle and jaw function	15 (27.8)	6 (11.1)	0.118 ^a
(5) Oral motor function	4 (7.4)	6 (11.1)	0.741 ^b
(6) Speech	42 (77.8)	15 (27.8)	0.005 ^a

NOT-S: nordic orofacial test-screening; UCLP: unilateral cleft lip and palate. ^aChi-squared test; ^bFisher's Exact test (comparison between groups)

Table 3 shows the comparison of the CPQ₈₋₁₀ scores between the groups. The mean overall scores for UCLP and the controls were 17.2 and 13.4 ($p = 0.132$), respectively. There was a significant difference between the groups only for the social well-being domain, with the UCLP group reporting, on average, more negative impacts than the controls (4.4 vs. 2.7, $p < 0.01$). However, the ES of 0.42 was only moderate and indicated that the difference between the groups was small in relation to the variability within the sample as a whole. Among the 10 items of social well-being domain, two were more reported by the UCLP group than the controls: being teased (0.9 vs. 0.2; $p < 0.01$) or asked about their condition (1.4 vs. 0.3; $p < 0.001$) by other children.

Table 4 shows the distribution of the responses to the global ratings of the CPQ₈₋₁₀ in both groups. There was no difference between the two groups in their global ratings of OH. The majority of children in the UCLP and control groups reported that the

health of their teeth and mouth was OK (37%) or very good (57.4%), respectively. There was a significant difference between the two groups in their ratings of the extent to which their oral or orofacial condition affected their life overall, with the control group perceiving it as somewhat better than the UCLP patients (48.1 vs. 20.4%; $p < 0.05$). The majority of children in the UCLP and controls groups reported that their condition had little (40.7%) or no effect (48.1%) on their life overall.

Correlations between NOT-S and CPQ₈₋₁₀ scores for each group are shown in Table 5. The NOT-S total scores were positively correlated with the CPQ₈₋₁₀ total and subscales scores for both groups, except for the oral symptoms in the UCLP group. There were positive correlations between the NOT-S interview scores and the CPQ₈₋₁₀ total and subscales scores for both groups. The NOT-S examination score was only significantly correlated with the social domain scores in the control group ($r = 0.28$, $p < 0.05$).

Table 3. Comparison of CPQ₈₋₁₀ overall and domain scores between groups.

Variable	Range of possible scores	UCLP group	Control group	p-value*
		Mean (SD)	Mean (SD)	
Overall scale	0–100	17.2 (13.0)	13.4 (12.1)	0.132
Oral symptoms	0–20	6.0 (2.8)	5.2 (3.8)	0.239
Functional limitations	0–20	3.7 (3.5)	2.4 (2.7)	0.060
Emotional well-being	0–20	3.1 (3.7)	3.2 (3.7)	0.798
Social well-being	0–40	4.4 (5.1)	2.7 (4.2)	0.010

CPQ: child perceptions questionnaire; UCLP: unilateral cleft lip and palate; SD: standard deviation. *p-value obtained from the Mann-Whitney test

Table 4. Distribution [n (%)] of responses to global ratings by group.

Variable	UCLP group	Control group	p-value*
When you think about your teeth or mouth, would you say that they are:			
Very good	16 (29.6)	31 (57.4)	0.097 ^a
Good	16 (29.6)	12 (22.2)	0.644 ^a
OK	20 (37.0)	10 (18.5)	0.157 ^a
Poor	2 (3.7)	1 (1.9)	1.000 ^b
How much do your teeth or mouth bother you in your everyday life?			
Not at all	11 (20.4)	26 (48.1)	0.050 ^a
A little bit	22 (40.7)	15 (27.8)	0.421 ^a
Some	19 (35.2)	13 (24.1)	0.464 ^a
A lot	2 (3.7)	0 (0.0)	0.496 ^b

UCLP: unilateral cleft lip and palate. ^aChi-squared test; ^bFisher's Exact test (comparison between groups)

Table 5. Correlation between NOT-S and CPQ₈₋₁₀ scores for the UCLP group (n = 54).

Variable		NOT-S scores	
UCLP group			
CPQ ₈₋₁₀ overall scale	0.38**	0.47***	0.02
Oral symptoms	0.25	0.35**	-0.05
Functional limitations	0.35**	0.47***	-0.02
Emotional well-being	0.37**	0.38**	0.11
Social well-being	0.29*	0.35**	0.02
Control group			
CPQ ₈₋₁₀ overall scale	0.46***	0.47***	0.26
Oral symptoms	0.37**	0.38**	0.14
Functional limitations	0.41**	0.44***	0.17
Emotional well-being	0.32*	0.36**	0.12
Social well-being	0.37**	0.35**	0.28*

NOT-S: nordic orofacial test-screening; UCLP: unilateral cleft lip and palate; CPQ: child perceptions questionnaire. *p < 0.05; **p < 0.01; ***p < 0.0001 (Spearman correlation test).

Discussion

This study was undertaken to evaluate the orofacial functions and OHRQoL in 54 pairs of children with and without UCLP matched by sex and age, using NOT-S and CPQ₈₋₁₀ measures, respectively. Both of the hypotheses tested were confirmed. That is, children with UCLP had poorer orofacial functions and more negative impacts on social well-being than controls. Further, the scores of both measures were correlated, suggesting that the more frequent their orofacial dysfunction, the greater the impairment of QoL in both populations. The strengths of this study include an *a priori* sample size calculation, as this is an essential item to reduce the probability of error²⁶, and matching controls by age and sex, which allowed for the homogeneous distribution of these variables in the groups. Another strength of this investigation was the use of standardized questionnaires and also the standardized assessment of the oral conditions by a single calibrated/trained examiner.

In this present study, mean NOT-S total and examination scores were higher in the UCLP group than in the control group, suggesting more clinical signs of orofacial dysfunctions in the former, especially related to the face at rest, facial expression, and speech. In relation to the face at rest domain, all children with UCLP had skeletal and/or soft tissue asymmetry, while 63% had a deviant lip position when opening the mouth. Nasolabial asymmetry is

a common finding in UCLP patients and in previous studies, using various methods to evaluate facial morphology, it has been shown that the cleft and non-cleft sides of the face differ.¹⁶

In relation to the facial expression domain, this domain measures the ability of the patient to pout and round the lips symmetrically, functions that depend on lip strength. In the present study, more than half of the patients (57.4%) could not pout or round their lips symmetrically. This is contrary to Van Lierde et al.'s¹⁸ findings of normal lip strength in children with unilateral UCLP. However, the comparison between the present findings and the above mentioned study should be interpreted with caution because they used different instruments: a screening instrument for clinical examination of orofacial dysfunction (NOT-S) and a hand-held device that measures the amount of pressure applied to a small pliable air-filled bulb in order to gage lip strength, tongue elevation strength, and tongue endurance (Iowa Oral Performance Instrument [IOPPI]).

In relation to the speech domain, the majority of the patients in this study presented with unclear speech with one or more indistinct sounds or abnormal nasality. Speech and voice specialists state that because of the anatomical constraints present in UCLP infants, even the earliest attempts at vocalization in the second and third months of life in these children are clearly different from infants without a cleft defect.²⁷ At about 6 months of age, a child without the defect

starts to produce its first frontal consonants, whereas the structural deformity in a child with UCLP makes such a task impossible and may give rise to glottal or pharyngeal articulations.

Moreover, one third of the UCLP patients reported abnormal breathing in contrast with only 7.4% of the controls. All reports were related to snoring when sleeping. Children with UCLP have a higher incidence of obstructive sleep apnea,²⁸ which is characterized by prolonged partial upper airway obstruction and/or an intermittent complete obstruction that disrupts both normal ventilation during sleep and normal neurophysiological sleep patterns.²⁹ The size of their pharyngeal airways has been shown to be smaller and their craniofacial dimensions differ from those of healthy controls.²⁹ The oropharyngeal musculature is also disrupted by the cleft, which impacts speech and swallowing, as well as adversely affecting the maintenance of airway patency, particularly during sleep.

The comparison between the present findings and other studies should be interpreted with caution because some studies used another OHRQoL questionnaire, different populations, and different methods. Broder and Wilson-Genderson³⁰ reported that craniofacial patients had greater negative impacts on social-emotional well-being and school environment than general pediatric or orthodontic patients. Other studies using CPQ found no statistically significant difference between 8–10-year-old children with UCLP and children with dental caries⁵ and healthy groups⁹ or found few differences in the OHRQoL of children aged 11 to 14 years with orofacial conditions compared with children with dental caries. Ward et al.¹⁰ found worse OHRQoL for the social-emotional well-being and functional well-being subscales among children with orofacial clefts compared with controls. In the present study, although the difference in the social well-being domain was statistically significant between the UCLP patients and controls, the ES suggested that the magnitude of the difference in the mean score for the two groups was moderate in relation to the variability among the study participants as a whole. Moreover, the mean difference between the groups was only 1.7 on a scale that could range from

0 to 40. Additional evidence that the higher scores of the CPQ₈₋₁₀ in the UCLP group may not be that important is to be found in the responses to the two global items. Overall, UCLP patients rated the health of their teeth and mouth similar to controls and frequently reported that their condition had little effect on their life overall (40.7%). This leads to the conclusion that although the UCLP group may encounter more challenges in daily life, their overall QoL is no different from that of healthy children. This confirms the results of an earlier study that used CPQ₁₁₋₁₄ to measure the outcomes of orofacial conditions and dental caries in children aged 11 to 14 years.³¹ The OHRQoL of the participants with cleft lip and/or palate was similar to that with dental decay. The present and above mentioned findings are consistent with the observation by Strauss³² in a literature review that, although appearance and speech may remain problematic for some people with orofacial conditions, the majority are “productive, contributing, happy, satisfied individuals.”

The correlation analysis between the NOT-S and CPQ₈₋₁₀ scores indicated an association between orofacial dysfunctions and worse OHRQoL in both groups in this study. However, clinical evidence of this relationship was observed only for the control group who presented a significant positive correlation between the NOT-S examination and the CPQ₈₋₁₀ social domain scores. The patient-reported outcome measures would ascertain the patient’s views of their symptoms, functional status, and HRQoL. However, assessing individuals with CLP can be difficult because congenital anomalies require long-term follow-up periods and serial assessment strategies to monitor treatment progress over time.³³ In this context, our results may help in the understanding of how eight- to ten-year-old non-syndromic children with clefts perceive their health, guiding the professionals to provide a more holistic treatment. It is important to recognize the limitations of this work in terms of the methodology and analytic strategies used. The lack of temporality limits our confidence in establishing the direction of associations. Notwithstanding, cross-sectional investigations are important tools to identify risk indicators to be included in further longitudinal

assessments. So, further research is needed to better explain the link between orofacial dysfunctions and OHRQoL in patients with UCLP.

Conclusions

Children with UCLP reported more orofacial dysfunctions and negative impacts on social well-being than controls. Moreover, the more frequent the

orofacial dysfunction, the greater the impairment of QoL in both populations.

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