Original Article

Impact of Oral Clefts on the Oral Health-Related quality of Life of Preschool Children and their Parents

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Abstract

Objective: To assess the oral health-related quality of life (OHRQoL) of preschool children with cleft lip and palate (CLP) and their relatives. **Materials and Methods:** In this cross-sectional study, 55 2–5-year-old children with the history of CLP were randomly selected from those referred to Shiraz Lip and Palate Cleft Research Center and treated with single-stage closure (Push back palatoplasty). Furthermore, same number of children with the same age who attended the Shiraz School of Dentistry for routine dental care were selected as control group using randomized sampling. Children's demographic data were obtained from their parents. Farsi version of the Early Childhood Oral Health Impact Scale (F-ECOHIS) was used for evaluating these children's QoL. **Results:** We found a significant difference in OHRQoL between children with CLP and children without CLP in the overall score of F-ECOHIS and all of subscales. In the impact on children subscale, the difference between these groups was remarkable in limitations' domain. As for difficulties faced by children, question on "difficulty in pronouncing words" had the highest average score. Furthermore, in impact on family, in parental distress domain, the difference between these groups was remarkable. For difficulties faced by family, financial impact got the highest average score. No significant difference was found between boys and girls with CLP in all subscales. While according to the score of total F-ECOHIS in unilateral and bilateral CLP children, there was statistically significant difference in these groups. **Conclusion:** Since oral clefts affect the QoL of children and their families even after the usual treatments, the implementation and maintenance of multidisciplinary interventional strategies are required for establishment of facial esthetics, oral function, and psychological support for such individuals.

Keywords: *Cleft lip, cleft palate, oral health, quality of life*

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Introduction

The World Health Organization definition of health as both the absence of disease and the presence of factors that promote physical, mental, and social well-being has led to a wider conceptualization of overall health which necessarily results in quality of life (QoL).[1] Given the extent of oral diseases, these are the main public health problems accounting for disability in every part of the world. The pain, suffering, and impairment of functions such as eating, chewing, smiling, communication, and the decreased QoL due to oral diseases mean that their effect on individuals and communities is remarkable.[2] Moreover, the successful consequences of medical treatments for children are not merely defined in terms of cure, repair, or remission but also in terms of extend to the maintenance or improvement of patients' QoL after treatment,[3] particularly for children with chronic health conditions,

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who need long-term treatment protocols such as children with craniofacial abnormalities.[4]

Oral clefts with prevalence of 9.92–10/10,000 live birth worldwide and Iran, respectively, are one of the most widespread birth defects.[5-7] These deformities have great influences on all aspects of both patients' health and their families' health. Oral clefts are responsible for a high burden of disease due to their complexity.

In addition to the esthetical defects, cleft palate deformities are associated with a large number of problems such as speech disorders, hearing deficits, chronic ear infections (serious otitis media), dental and palatal deformities, and psychosocial problems.[8]

Oral health-related QoL (OHRQoL) was expanded to help to assess the physical and psychosocial effect of oral health. There are attempts to quantify the extent to which dental and oral disorders interfere with daily life and well-being together with the outcomes of clinical care like the effectiveness of treatment interventions.[9] Speech and esthetic concerns have appeared to be crucial factors influencing the health-related QoL after repairing surgeries in cleft lip and palate (CLP) children.[4,10] Moreover, significant psychological and social burdens have been reported in relation to orofacial clefts. Psychological adjustment tends to be challenging for children with orofacial clefts in view of esthetic concerns, speech and hearing disabilities, and difficulty in acquiring the social skills essential for adjustment.[10]

Studies have developed and tested different OHRQoL questionnaires for children aged from 6 years or older.[11-13] For younger children, on the other hand, this kind of research is limited. Therefore, the Early Childhood Oral Health Impact Scale (ECOHIS)[14] was developed to assess the burden of oral diseases and its treatment among preschool children. Furthermore, it is intended to investigate the perception of parents, as the target population is composed of children at preschool age, who often times pose some challenges while answers to proposed questions are elicited.[15]

Scarpelli *et al*. in their study evaluated the QoL of preschool children from different social classes regarding the presence of early childhood caries, tooth trauma, malocclusion, the developmental enamel defects, and decayed, missing, and filled teeth using ECOHIS associated with a socioeconomic questionnaire. They found that dental caries experience was the only normative criteria with a negative impact on OHRQoL.[16] Sousa *et al*. applied Brazilian version of ECOHIS to evaluate the impact of malocclusions on the QoL of preschoolers and their families considering the esthetic and functional consequences of this oral problem and illustrated that malocclusion was not associated with a negative impact on OHRQoL.[17] Gomes *et al*. assessed the QoL of 3–5-year-old children, through ECOHIS, regarding caries, tooth trauma, and malocclusion from the physical and psychological consequences of these conditions. According to their study, cavitated lesions and traumatic dental injury exerted an impact on OHRQoL of the preschool children and their families.[18]

Few studies revealed that the CLP negatively impacted on OHRQoL of children and their parents in different ages using ECOHIS.[15,19] Farsi version of the ECOHIS (F-ECOHIS) is the Farsi version of ECOHIS and likewise has high level of sensitivity and accuracy and has been used in several studies.[20] Although this questionnaire is proved to have a high level of sensitivity and accuracy, no actual study had ever tried to evaluate QoL in 2–5-year-old patients with oral clefts in comparison to control groups without oral clefts, using F-ECOHIS. Therefore, the current study, to the best of the authors' knowledge, is the first study to evaluate the QoL of 2–5-year-old patients with CLP applying F-ECOHIS and compare it with peer children without CLP. As research in the field of QoL in these children is so important, this study is focused on the QoL of patients with CLP, considering the burden of disease in CLP children and impact of this condition on the general health and wellbeing of the children and their families.

Materials and Methods

This cross-sectional study was approved by the Ethics Committee of Shiraz University of Medical Sciences (ref. no. 95-01-03-11604), and the informed consent was taken from all participants. Fifty-five 2–5-year-old children with the history of CLP were randomly selected from those referred to Shiraz Lip and Palate Cleft Research Center and treated with single-stage closure (Push back palatoplasty). Furthermore, same number of children with the same age who attended the Shiraz School of Dentistry for routine dental care were selected as control group, using randomized sampling. According to medical history, all children with systematic and chronic disease and syndromic CLP patients were excluded from the study.

The children's OHRQoL was evaluated through applying an oral health-related questionnaire: the Farsi version of the ECOHIS (F-ECOHIS).[20] It considers the child's entire lifetime experience of dental disease and treatment in parent's responses. Indeed, it evaluates the perception of parents on OHRQoL of their 2–5-year-old children. The F-ECOHIS questionnaire contains 13 questions, 9 included in the impact on children section and 4 in the impact on the family

section. The impact on children section was divided into 4 subclasses (domains): symptoms (related to pain); limitations (including difficulty in drinking, difficulty in eating, difficulty in pronunciation, and missing daycare); psychological (difficulty in sleeping and getting annoyed); and self-image (avoid smiling and void talking) aspects. The impact on family section was divided into two subclasses (domains): parental distress (getting annoyed and feeling guilty) and family function (missing work and financial impact). Response options can be 0 = never; 1 = hardly ever; 2 = occasionally; 3 = often; 4 = very often; and 5 = don't know.[20] The score for the child and family sections has a possible range from 0 to 36 and from 0 to 16. Higher scores indicate a more negative impact on the OHRQoL or vice-versa.

Data were described using mean (± standard deviation [SD]). To assess the normality of distribution of the values, a Kolmogorov–Smirnov test was used. Student's *t*-test was employed to compare mean ECOHIS scores between the groups. Data were recorded in a Statistical Program Software database - SPSS 18.0 (SPSS Inc., Chicago, IL, USA). $P < 0.05$ was considered statistically significant.

Results

A total of 102 questionnaires were collected and used for the final analysis. Fifty-five (57% boys and 42.6% girls) were caregivers of children with CLP who were referred to the Shiraz Lip and Palate Cleft Research Center. They were either with unilateral (61%) or bilateral (38%) CLP anomaly. Forty-seven (50.9% boys and 49.1% girls) were caregivers of children without CLP. Patients were in the age range of 2–5 years, which was the required range for ECOHIS.

All survey participants reported some impact of CLP on QoL of children and their family. According to the parents' perception on the OHRQoL of children with and without clefts, the statistical analysis of the questionnaire showed a statistically significant difference between groups with higher impact of the cleft on the OHRQoL [Table 1]. As shown in Table 1, in impact on children section, there was statistically significant difference between children with CLP and without CLP. As for difficulties faced by children, in "limitations" domain, the difference was remarkable and of course, statistically significant ($P < 0.001$). In this domain, question on "difficulty in pronouncing words" had the high average score of 4.45 (SD: 0.91), and its difference with the control group was very significant [Table 1]. Furthermore, in impact on the family section, we found statistically significant differences between these groups, and the difference was remarkable in parental distress domain. Moreover, for difficulties faced by family, financial impact had the highest average score of 4.56 (SD: 0.50). All domains of F-ECOHIS questionnaire were statistically significant in both groups ($P < 0.001$).

Table 1: Mean scores in patients with and without oral clefts

F-ECOHIS	Mean±SD		*P*
	Children without CLP (*n*=47)	Children with CLP (*n*=55)	
Domains			
Subscale impact on the child	15.85±5.16	39.14±2.16	<0.001
Symptoms	2.76±0.75	4.83±0.37	<0.001
Limitations	7.89±2.29	18.27±1.87	<0.001
Psychological	2.85±1.54	8.03±1.23	<0.001
Self-image	2.34±0.86	8.00±1.80	<0.001
Subscale impact on the family	6.55±2.78	18.16±1.52	<0.001
Parental distress	3.27±1.37	9.20±0.98	<0.001
Family function	3.27±1.44	8.96±0.66	<0.001
Total F-ECOHIS	22.40±1.65	57.30±3.22	<0.001

SD=Standard deviation; CLP=Cleft lip and palate; F-ECOHIS=Farsi version of the Early Childhood Oral Health Impact Scale

Table 2: Mean scores of cleft lip and palate patients according to their sex

	Male	Female	*P*
Domains			
Subscale impact on the child	39.14±2.31	39.14±2.04	0.993
Symptoms	4.81±0.39	4.85±0.35	0.678
Limitations	18.33±1.96	18.21±1.81	0.816
Psychological	8.07±1.26	8.00±1.21	0.826
Self-image	7.92±1.79	8.07±1.84	0.768
Subscale impact on the family	18.29±1.48	18.03±1.57	0.531
Parental anguish	9.25±0.98	9.14±1.00	0.667
Family function	9.03±0.64	8.89±0.68	0.427
Total F-ECOHIS	57.43±3.39	57.17±3.11	0.713

F-ECOHIS=Farsi version of the Early Childhood Oral Health Impact Scale

Mean scores of CLP patients according to their sex are presented in Table 2. No significant difference was found between male and female patients with CLP regarding mean scores of F-ECOHIS and its subclasses. As shown in Table 3, according to the score of total F-ECOHIS in unilateral and bilateral CLP children, there was statistically significant difference in these groups ($P < 0.001$). Moreover, in subscale impact on the child and family, there was statistically significant difference between these groups ($P < 0.001$). Domains of limitations, psychological, and self-image in the subscale impact on the child and parental anguish in the subscale impact on the family were statistically significant ($P < 0.001$).

Table 3: Mean scores of cleft lip and palate patients according to their type of cleft

ECOHIS	Mean±SD		*P*
	Unilateral CLP (*n*=29)	Bilateral CLP (*n*=26)	
Domains			
Subscale impact on the child	38.58±2.23	40.47±1.74	0.014
Symptoms	4.76±0.43	4.95±0.21	0.070
Limitations	17.70±1.94	19.19±1.32	0.001
Psychological	7.64±1.27	8.66±0.85	0.001
Self-image	8.47±1.84	7.23±1.48	0.009
Subscale impact on the family	17.73±1.58	18.85±1.15	0.004
Parental anguish	8.88±1.00	9.71±0.71	0.001
Family function	8.85±0.70	9.14±0.57	0.117
Total F-ECOHIS	56.02±3.18	58.33±2.81	0.009

SD=Standard deviation; CLP=Cleft lip and palate; ECOHIS=Early Childhood Oral Health Impact Scale; F-ECOHIS=Farsi version of the Early Childhood Oral Health Impact Scale

Discussion

We found a significant difference in OHRQoL between children with CLP and children without CLP in the overall score of F-ECOHIS and all of subscales. In the impact on children subscale, the difference between these groups was remarkable in limitations domain. As for difficulties faced by children, question on "difficulty in pronouncing words" had the highest average score. Furthermore, in impact on family, in parental distress domain, the difference between these groups was remarkable. For difficulties faced by family, financial impact got the highest average score. There were no significant differences between boys and girls in children with CLP. While statistically significant difference was found between unilateral and bilateral CLP children, according to the score of total F-ECOHIS.

The impacts on children were statistically more than the impacts on the family in the present study. In the other studies, parents reported a greater impact on QoL of children over the QoL of families, agreeing with similar study although these studies investigated other disturbing oral conditions.[15,21,22]

In the previous studies of OHRQoL using ECOHIS,[22-24] differences in subscales related to pain due to dental caries and dental trauma were the most pronounced ones. In the present study, subscale related to pain obtained the lowest score. The most relevant issue was difficulty in pronouncing words, and the difference between affected and control group was significant. In addition to the difficulties of speech, higher hearing problems and ear infections in individuals affected by oral cleft was remarkable.[15]

It is noteworthy that the articulation errors in sound production may be the characteristic of the child's normal development in the investigated age. Nevertheless, it has been shown that the tendency of the impact of oral clefts is more related to compensatory errors in sound production, and it is known that a child carrier with oral clefts faces other limitations in the learning process of speech. A cleft palate during the development of motor control of speech and associated sensorimotor systems changes the learning of speech. Errors seem to appear more when surgical palatal repair is not performed ideal. This is also approved by neuropsychological assessment survey of children with clefts that one of the functions reported to be affected by malformation is the cognitive-linguistic function.[15,25] In a survey that assessed satisfaction about esthetics and function of individuals with oral clefts, even after receiving orthodontic treatment, speech impairment was reported as a reduced satisfaction parameter.[26]

The parameters investigated by this questionnaire reveal the negative feelings that the child's malformation could generate for the families which can be explained by parental distress and family functions. Similarly, another study that evaluated CLP children demonstrated that difficulties in the daily life of the child-family binomial for various issues reported by the family, such as fear and uncertainty of what might happen to the child, match the scale of negative feelings.[27]

Recent studies demonstrate that reports of children on OHRQoL are reliable and valid. Instruments developed to measure OHRQoL of children should also assess the impact of these problems on the family's QoL because they are inseparable factors.[18,28] The assessment of OHRQoL of the child reflects on the parents' perception toward their own oral health, thus improving the communication between children, parents, and dental health professionals.[29] Considering the F-ECOHIS scores, there was no significant differences in CLP children according their sex. While other studies found significant differences between boys and girls with CLP on the subscale, emotional well-being, and peer interaction in children older than 8 years old.[30,31] This indicates that QoL of girls was more affected by oral health as children growing up. Furthermore, similar to our result, another study indicated that bilateral CLP group which included more severe cases had worse OHRQoL than the other types of cleft.[30] As bilateral CLP has more complications and has great impact on domains of limitations, psychological, self-image, and parental anguish, QoL is greatly affected in these children. This may explain the fact that the impact of esthetic and functional impairment on QoL in different sexes is age dependent and may be revealed in older age with more peer interactions and social activities.

As a limitation, treatment outcome satisfaction was not evaluated in the current study. It is important to understand that although CLP patients would inevitably undergo surgical treatment, their experience of the difficulties and complications of this kind of treatment would be different. Since treatment outcome can affect OHRQoL, as a clinical relevance of this study, we suggest that researchers consider evaluating the subjective treatment outcome of surgeries in CLP patients in future similar studies.

Furthermore, although ECOHIS is a valid questionnaire for evaluating OHRQoL in CLP patients, in-depth psychological assessment of CLP patients is recommended to identify main concerns of the patients and their parents. This would help clinicians to detect unrealistic treatment outcome expectations and psychological and cognitive problems of CLP patients and finally to evaluate the treatment effects.

As it was mentioned, difficulty in pronouncing words was the worst problem that these children face, even years after surgical treatment. Therefore, rehabilitation strategies should be especially programmed for speech therapy to improve these children's QOL.

Conclusion

Findings of this study showed that the CLP children had a remarkable higher F-ECOHIS score, representing much worse OHRQoL than their peers. The "limitation" domain of F-ECOHIS had the highest expression on children. Inside this domain, question on "difficulty in pronouncing words" had the highest average score. Regarding the section on "impact on the family," the "parental distress" domain had the highest average score, and in this domain, financial impact was more prominent.

No significant difference was found between boys and girls with CLP. While according to the score of total F-ECOHIS in unilateral and bilateral CLP children, there was statistically significant difference in these groups.

Since the presence of clefts impacts the QoL of children and their families, the implementation and maintenance of multidisciplinary interventional strategies such as neonatal orthopedic treatments and interventions before speech therapy are required for the reestablishment of esthetics, function, and psychological support for such individuals. Furthermore, interventions that reduce parenting stress and enhance children's emotion regulation strategies may decrease the risk for later psychological problems in this population.

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Conflicts of interest

There are no conflicts of interest.

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