Article

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Abstract: Patients with Cleft Lip Palate (CLP) require comprehensive treatment, so the people who take care of them must constantly provide care and support, along with continuous care and support from their caregivers, often extending until youth. The purpose was to compare the Quality of Life (QoL) of caregivers of children with CLP with those without the condition. A cross-sectional paired study was conducted at the Dental Clinic of the Escuela Nacional de Estudios Superiores (ENES León), National Autonomous University of Mexico (UNAM), from May to December 2021. Primary caregivers of patients with CLP (n=70) were included as well as those of the patients without CLP (n=70) of the same institution. Sample size was calculated considering an OR=2.5, establishing a convenience selection. Caregivers answered the WHOQoL Bref instrument to assess QoL. Of the total sample analyzed, 88.6% of the caregivers were female ($p>0.05$). Caregivers of patients with CLP reported poor QoL (64.8%), compared to caregivers in the control group ($p<0.05$). The multivariate analysis showed that having a child with CLP increases stress and the time dedicated to caregiving, thereby increasing the likelihood of poor QoL ($p<0.05$). QoL was more adversely affected among caregivers of CLP patients. It is essential to provide comprehensive support not only to CLP patients, but also to the caregivers, since their well-being significantly impacts the QoL of the patients.

Keywords: cleft lip; cleft palate; caregivers; quality of life

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1. Introduction

Cleft Lip and Palate (CLP) is an abnormal congenital cleft that strongly affects the oral cavity and related structures [1]. Its multifactorial etiology includes genetic and epigenetic factors, especially environmental factors [1]. The worldwide incidence of CLP is 1:600/800 live births (1.42:1000), while isolated cleft palate (CPO) occurs in approximately 1 in 2000 births. Males are more affected than females, in a 2:1 ratio [2].

In Mexico, a prevalence of CPO/CLP of 5.3:10000 births have been reported, mostly in males (6.3:10 000 births) compared to females (4.2 per 10000 births) [3]. Likewise, in Guanajuato, Mexico, the observed prevalence was above the national rate of 6.2 per 10,000 births from 2008 to 2014 [3].

CLP strongly impacts both functionality and aesthetics, profoundly affecting patients' psychological and social aspects. Moreover, CLP influences not only the patient, but also their family members [4]. It has been reported that the the period when parents are first informed about their child's condition is particularly challenging, often marked by confusion, anguish, guilt, loss of control, impotence, stress, anxiety, and depression as they go through a period of mourning [5,6].

In addition, the costs of health services for the correction and rehabilitation of CLP generate a significant financial burden, which is, on average, eight times higher than that for children without this condition [7]. Furthermore, primary caregivers of children with CLP must frequently attend medical appointments, resulting in absences from work and the need to change or postpone previously planned activities [7]. These factors can cause an overload of work experience that reduces quality of life, leading to illness or stress-related conditions [8]. Thus, the well-being of children with CLP caregivers can have substantial repercussions on their quality of life [9,10].

However, since the perception of quality of life is self-perceived and dependent on particular social and demographic conditions, it is necessary to to gather extensive data on various situations to better understand patients and their caregivers. Therefore, the purpose of the present study is to compare the quality of life of primary caregivers of children with CLP compared to caregivers of children without this condition at the the dental clinics of the ENES León UNAM, during the year 2021.

2. Materials and Methods

This study was approved by the Research and Ethics Commission of the ENES León UNAM (CEI_21_04_S15). ENES León offers a university program that provides comprehensive care for patients with orofacial clefts named "TiENES que sonreir, UNAMos esfuerzos". Caregivers of patients in this program were invited to participate in the study. After explaining the objectives, they were extended a voluntary invitation to join. Their participation was formalized through the signing of an informed consent form.

The present study was a matched cross-sectional study. The study population consisted of 300 primary caregivers of patients with CLP who attended the program mentioned above, while the control population consisted of 150 caregivers of children who participated in the pediatric dentistry clinic of the same institution. The sample size was calculated under the following assumptions: confidence = 95.0%, power = 80.0%, probability of exposure in controls = 0.35, OR= 2.5, and Non-response Bias of 0%. Thus, a sample size of 70 mother-child pairs per group was determined. The pairs were chosen by convenience.

Caregivers were included if they were living in the same house as the patient and provided primary care for them for at least 8 hours per day. Those with visual or hearing problems that prevented them from answering the questionnaire were excluded, and those who did not answer the questionnaire in its entirety were eliminated. The children's data were obtained from clinical records, excluding those with incomplete or illegible information. The age of the children was used as the matching criterion.

After obtaining the signed consent form, a questionnaire on sociodemographic data and QoL was administered. The latter was assessed using the World Health Organization Quality of Live (WHOQOL-Bref) instrument, validated in Spanish. WHOQOL-Bref consists of 26 Likert-type questions, which were rated on a scale from 1 to 5, where 1= Never, 2= Almost never, 3= Sometimes, 4= Frequently and 5= Almost always. The score obtained ranges from 26 to 130. The higher the score, the better the quality of life.

The WHOQOL-Bref encompasses four domains: aspects of physical health and ability to engage in different activities (7 items); psychological health related to body image and appearance (6 items); social relationships and social support resources (3 items); and environment involving the material and health care resources available (8 items). It also contains questions on the overall perception of quality of life and general health satisfaction. It should be mentioned that this variable was categorized into two according to the median of the quality-of-life scale. For statistical analysis, descriptive measures were taken for the socio-demographic variables, frequencies for qualitative variables, and measures of central tendency and dispersion for quantitative variables. Measures of central tendency and dispersion were obtained for the caregivers' quality of life. Bivariate chi-square analyses were performed to verify differences in quality of life between the study groups. Finally, a binary regression-logistic model was used, entering variables that were significant in the bivariate analysis with a p-value ≤ 0.20.

Data from clinical records were used to evaluate clinical variables such as the type of cleft, affected site, and structures involved.

3. Results

One hundred forty caregivers participated in the study; 88.6% were female (mean age 35.34 ±9.4), and 11.4% were male (mean age 36.06; ±4.9). No statistically significant difference was observed according to the sex distribution (p>0.05).

The results obtained when comparing the sociodemographic variables between the study groups are shown in Table 1. It can be observed that the caregivers in the control group had more schooling than the study group since 54.3% of them had a high school level or higher (p=0.003).

Table 1. Socio-demographic data of caregivers of patients attending the dental clinic of ENES León.

		CLP patient caregivers		**Non-CLP patient caregiver**		**Total**		***p**
		n	%	n	%	n	%	
Gender	*Female*	64	91.4	60	85.7	124	88.6	0.288
	Male	6	8.6	10	14.3	16	11.4	
	Total	70	100	70	100	140	100	
Scholarship	*Primary school*	14	20	9	12.9	23	16.4	0.003
	Middle school	38	54.3	23	32.9	61	43.6	
	High school and upper	18	25.7	38	54.3	56	40	
	Total	70	100	70	100	140	100	
Occupation	*Employee*	21	30	27	48.6	48	34.3	0.016
	Unemployed	44	62.9	29	31.4	73	52.1	
	Professional	1	1.4	9	12.9	10	7.1	
	Other	4	5.7	5	7.1	9	6.4	
	Total	70	100	70	100	140	100	
Marital Status	*Singles/Free Union*	24	34.3	12	17.1	36	25.7	0.043
	Married	43	61.4	51	73.9	94	67.1	
	Divorced/Widowed	3	4.3	7	10	10	7.1	
	Total	70	100	70	100	140	100	
Monthly Economic Income	*Low*	19	27.1	14	20	33	23.6	

	Middle	49	70	53	75.7	102	72.9	0.573
	High	2	2.9	3	4.3	5	3.6	
	Total	70	100	70	100	140	100	

CLP= Cleft Lip Palate; *=chi-square.

The mean age of all children of both study groups was 8.8 ±3.4 years, 9.30 ±3.3 years for girls, and 8.36 ±3.6 for boys. Table 2 shows that 60% of the patients with CLP were boys, while 51.4% of the patients without CLP were girls. No statistical difference was observed in sex distribution between the two groups (p=0.175). Similarly, 80% of the mothers fulfilled the role of caregiver, regardless of the study group, 84.3% in the CLP group, and 75.7% in the non-CLP group. No statistically significant difference was observed according to the caregiver's relationship by group (p=0.39) (Table 2)

Table 2. Socio-demographic data of patients attending the dental clinic of ENES León.

		CLP patient		**Non-CLP patient**		**Total**		
		n	%	n	%	n	%	***p**
Gender	*Female*	28	40	36	51.4	64	45.7	0.175
	Male	42	60	34	48.6	76	54.3	
	Total	70	100	70	100	140	100	
Relationship	*Father*	6	8.6	11	15.7	17	12.1	0.390
	Mother	59	84.3	53	75.7	112	80	
	Other	5	7.1	6	8.6	11	7.9	
	Total	70	100	70	100	140	100	

CLP= Cleft Lip Palate; *=chi-square.

Regarding the clinical characteristics of patients with CLP, 44% presented with unilateral cleft lip and palate, 24% with isolated cleft palate, and 20% with bilateral cleft lip palate. The total number of cases is presented in Table 3. 87% of the patients reported having undergone one to three surgeries, 7% reported having undergone four or more surgeries, and only 6% had not undergone any surgery at the time of the present study.

Table 3. Distribution by cleft type and number of surgeries received of CLP patients attending the program "TiENES que sonreír, UNAMos esfuerzos" ENES, León.

		Frequency	
		n	%
Cleft type	*Unilateral cleft lip and palate*	31	44
	Isolated cleft palate	17	24
	Bilateral cleft lip and palate	14	20
	Unilateral cleft lip	7	10
	Bilateral cleft lip	1	2
Number of surgeries	*None*	4	6
	One to three surgeries	61	87
	Four or more surgeries	5	7

Table 4 shows that mothers spent more hours per day caring for the child with CLP with an average of 17.231 (SD±7.751) hours per day, compared to fathers who reported an average of 12.061 (SD±8.322) hours per day (p=0.014).

Table 4. Time spent by caregivers on patient care at the Dental Clinic of ENES León.

		n	**Mean**	**SD**	***p**
How many hours a day do you spend caring for your relative?	*Female*	124	17.23	7.751	0.014
	Male	16	12.06	8.322	
	Total	140			

*=*t*-student test.

Regarding the quality of life of female caregivers, 45 of the 65.2% of caregivers of children without CLP reported a good quality of life, while 46 (64.8%) of caregivers of children with CLP reported a poor quality of life (p=0.0001), as seen in Table 5.

Table 5. Quality of life of caregivers of patients attending the dental clinic of ENES León.

		Good quality of life		**Poor quality of life**		**Total**		***p**
		n	%	n	%	n	%	
Caregiver of patient with/without CLP	*Caregivers of patients without CLP*	45	65.2	25	35.2	70	50	0.0001
	Caregivers of patients with CLP	24	34.8	46	64.8	70	50	
	Total	69	100	71	100	140	100	

CLP= Cleft Lip Palate; *=chi-square.

No relationship was observed between reported quality of life, whether categorized as good or poor, and the number of surgeries undergone by their child (p=0.893).

The analysis of the four domains of the WHOQoL Bref instrument showed that the physical health domain showed a significant difference between the two groups. The group of caregivers of patients without CLP reported a higher score on the physical well-being domain than caregivers of children with CLP (66.326 vs. 59.234, respectively; p 0.001). Similarly, caregivers of patients without CLP reported higher scores in the environmental domain (p = 0.005).

Table 6. Quality of life across dimensions of the WHOQoL Brief instrument, as applied to caregivers of patients attending the dental clinic of ENES León.

		n	**Mean**	**SD**	***p**
Physical Health	*Non-CLP patient caregivers*	70	66.326	15.496	0.001
	CLP patient caregivers	70	59.234	8.884	
	Total	140			

Psychological Health	*Non-CLP patient caregivers*	70	64.345	16.524	0.233
	CLP patient caregivers	70	61.071	15.828	
	Total	140			
Social relationships	*Non-CLP patient caregivers*	70	57.857	22.019	0.364
	CLP patient caregivers	70	55.00	14.286	
	Total	140			
Enviroment	*Non-CLP patient caregivers*	70	57.053	16.838	0.005
	CLP patient caregivers	70	50.178	11.133	
	Total	140			

CLP= Cleft Lip Palate; *= *t*- Student Test.

4. Discussion

The primary caregivers of children with dysmorphology, including CLP, play a crucial role as both psychological and financial supporters of the individuals under their care. Therefore, any imbalance in emotional and mental stability can affect the patient's behavior [11]. The psychological status and the quality of life of the parents of patients with orofacial clefts, mainly the primary caregivers, are factors that impact. In the present study, a comprehensive assessment of quality of life was conducted using the WHOQoL Bref instrument that allowed the analysis of physical, psychological, and social aspects of the primary caregivers of patients, both with and without CLP.

Our results reveal that mothers predominantly assumed the role of primary caregivers, regardless of the study group, which is consistent with previous reports of scientific literature [12,13], according to which they decicated 17 to 24 hours a day to their care. Ribiero et al. [12] pointed out that a close relationship between mother and child is paramount for rehabilitating patients with CLP. In recent years, it has been reported [14] that fathers are also involved in childcare, although mothers typically remain more actively engaged in the primary caregiving role.

Given the above, it becomes evident that caring for children with CLP affects the primary caregivers' lives and their immediate family environment in different ways. On the one hand, the hours invested in caring for infants can condition the entry of caregivers into the labor market, leading to school dropout and acceptance of part-time jobs and, consequently, leading to lower incomes [15]. These findings are consistent with our results, as they revealed that mothers, who were the primary caregivers of CLP patients, often had lower levels of education and were frequently unemployed, contrasting with the control group where unemployment rates were lower. This disparity may stem from the demanding nature of caring for a child with CLP, requiring round-the-clock availability to accompany the patient during treatment, particularly in the early years of life. The care of children with special healthcare needs can profoundly affect labor participation, which is directly proportional to the additional healthcare needs of the child [16]. Indeed, a study found that 80% of parents made employment decisions based directly on their child's health status, with one-third eventually ceasing work to care full-time for their child with special healthcare needs [17]. On the other hand, medical care for CLP patients often requires long-term commitments. While children without special health care needs gradually become more independent, caregivers of cleft patients may face longer-term care responsibilities, limiting or even preventing their return to regular employment [16].

Regarding the quality of life reported by caregivers, our results show a significant difference between the two groups. Caregivers of patients with CLP exhibited poorer quality of life than the control group. These findings are consistent with the scientific literature, which indicates that quality of life is better in caregivers of patients without clefting [18,19]. Specifically, families with a member who has CLP report significantly lower quality of life in the domains of physical, social, and psychological areas compared to control group families, especially as the patient grows towards adolescence [18]. As proposed by Hatzmann et al., caregivers of chronically ill children have a low quality of life and are at risk of further decline [20].

In our study, caregivers who reported worse quality of life were those caring for patients who had undergone one to three surgeries and those who had patients between 7 and 11 years of age. Although children with CLP have completed lip and palate reconstructive surgeries before this age, aesthetic rehabilitations, including dental, orthodontic, or orthognathic treatments and speech development interventions, will continue during adolescence [18]. However, a recent study found no statistical difference in the total score and the four dimensions of quality of life among caregivers of patients with cleft lip and palate [11]. The quality of life of a patient's parents is closely related to the patient's physical and psychological characteristics [11].

Our results indicated that caregivers with lower educational levels had a worse quality of life than those who had high school or upper academic degrees, and those caregivers who were unemployed reported a poorer quality of life than caregivers who were employed professionals. Previous studies have identified cleft type as a significant factor associated with the quality of life of caregivers of patients with CLP [13,19]. Specifically, families with children who have isolated cleft palate tend to have a better quality of life than those with cleft lip/palate or cleft lip [19]. However, in our study, cleft type was not related the quality of life of caregivers.

The binary logistic regression model showed that caregivers of patients with CLP were 3.2 times more likely to report a poor quality of life.

5. Conclusions

- Self-reported quality of life was lower among caregivers of patients with CLP.
- The presence/absence of CLP and the presence of stress were significantly associated with female caregivers' quality of life.

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: The data sets used and/or analysed during the current study are available from the corresponding author on reasonable request.

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Conflicts of Interest: The authors declare no conflict of interest.

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