

Measuring the Impact of Quality of Life of Children Treated for Orofacial Clefts: A Case-Control Study

Tannure PN*/ Soares FMM**/ K uchler EC***/ Motta LG**** / Costa MC*****/
Granjeiro JM *****

Objective: The aim of this study was to assess the quality of life (QoL) of children previously treated for cleft lip and/or palate (CL/P) and compare with non-cleft children. **Method:** A case-control study with 70 children between 5 and 12 years old was carried out. The case group consisted of 35 individuals previously treated for non-syndromic CL/P and presently receiving assessment at a rehabilitation hospital in Brazil. The children had received primary surgical treatment for CL/P reconstruction during early childhood. The control group consisted of 35 healthy children selected to ensure close similarity to the cleft group in age, gender and socio-economic status. QoL was measured using the AUQEI questionnaire. **Results:** Cleft lip and palate had no significant influence on the QoL in children ($p=0.44$). A higher percentage of the cleft lip and palate group of children reported a lower QoL than the cleft lip or cleft palate groups. Gender had no significant difference on the quality of life in CL/P children ($p=0.2$) and in control group ($p=1.0$). **Conclusion:** The QoL in children with CL/P was found to be similar to the non-cleft group. Our results confirm that clefts repaired during earlier childhood associated with a health care program, including psychological support, is beneficial for CL/P children.

Keywords: cleft lip; cleft lip and or palate; cleft palate; child; quality of life

INTRODUCTION

Cleft lip and/or palate are major structural birth defects that represent a significant public health burden. Roughly 1 in every 500 to 1000 births worldwide is affected¹ with concomitant lifelong morbidity.² The condition requires multiple surgical procedures from birth to maturity and frequent outpatient attendances. Many children with cleft lip and palates suffer impaired facial growth, dental anomalies, speech disorders, poor hearing, and difficulties in psychological wellbeing and social relationships.³

Depending on the type of cleft, children with oral clefts are stigmatized either because of their appearance (cleft lip: CL) or speech (cleft palate: CP) or both (cleft lip and palate: CLP) and therefore are probably particularly at risk to develop an altered social emotional functioning during early childhood.⁴ Measuring the quality of life (QoL) in children can be difficult, but a better understanding of how individual components correspond to overall measures of quality of life could help to improve the quality of life for individuals with oral clefts.⁵

The Autoquestionnaire Qualit e de Vie Infant Image (AUQEI) is a quality of life assessment questionnaire based on child (4-12 years-old) satisfaction, visualized and developed in France by Manificat and Dazort.⁶ The AUQEI has 26 items related to function (eg: meals), family (eg: parents), leisure (eg: vacation), autonomy (eg: alone) and others (eg: in hospital). It explores the view of the child or adolescent on those items through 4 different faces (very sad to very happy) using a visual analogue scale. Each item is then given a value from 0 to 3, so that the whole questionnaire may score from 0 to 78. Response levels are measured by checking faces expressing different emotional states. The AUQEI has been validated for Brazil and a total score below 48 was considered a very low quality of life.⁷ Previous studies have used the AUQEI to evaluate the quality of life of children after kidney or liver transplantations,⁸ mouth-breathing children,⁹ children with sleep bruxism¹⁰ and children and adolescents with spinal muscular atrophy.¹¹

The quality of life for cleft children is a priority for future public health research on CL/P.⁵ Recently controversial reports¹²⁻¹⁶ on the QoL of children with oral clefts have impaired a clear statement concerning the quality of life of treated children. In addition, patient-reported outcomes in CL/P treatment are critical for patient care. The aim of this study is to investigate if Brazilian children

* Patricia Nivoloni Tannure, Department of Pediatric Dentistry and Orthodontics, School of Dentistry, Federal University of Rio de Janeiro, RJ, Brazil; Pediatric Dentistry Discipline, School of Dentistry, Veiga de Almeida University.

** Fernanda Martins Moreno Soares, Department of Pediatric Dentistry and Orthodontics, School of Dentistry, Federal University of Rio de Janeiro.

*** Erika Calvano K uchler, Clinical Research, Fluminense Federal University, Niter oi.

**** Luise Gomes da Motta, 4Department of Dental Materials and Occlusion, Fluminense Federal University.

***** Marcelo de Castro Costa, Department of Pediatric Dentistry and Orthodontics, School of Dentistry, Federal University of Rio de Janeiro.

***** Jose Mauro Granjeiro, Unit of Clinical Research and Senior Researcher, National Institute of Metrology, Quality and Technology, Head of Bioengineering Program, and Head of Cell Therapy Center, Unit of Clinical Research, Fluminense Federal University, Niter oi, Rio de Janeiro.

Send all correspondence to: Professor Patricia Nivoloni Tannure, Pediatric Dentistry Discipline, School of Dentistry, Veiga de Almeida University, RJ-Brazil, Rua Ibituruna, 108 - Maracan  Rio de Janeiro - RJ, 20271-020

E-mail: pntannure@gmail.com

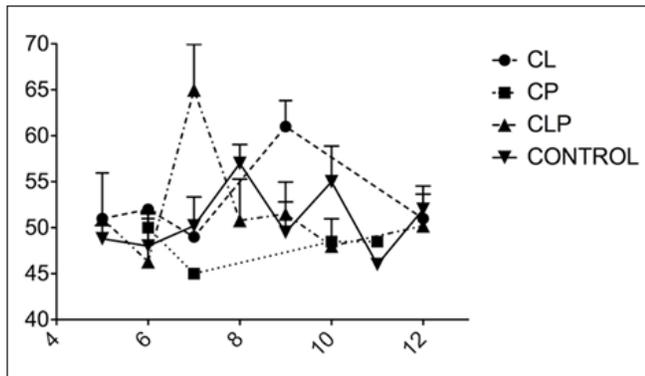


Figure 1. Relationship of age (5-12 years) and cleft type to mean AUQUEI scores and standard deviation

previously treated for oral clefts experience psychosocial distress and a reduction in their QoL.

MATERIALS AND METHOD

A case-control study design with 70 children aged between 5 and 12 years old was carried out. The case group consisted of 35 individuals previously treated for CL/P and who were receiving assessment at a public reference hospital for cleft rehabilitation in Rio de Janeiro Brazil. This study was carried out between January and July 2009. Children with clinically or genetically diagnosed syndromic oral clefts were excluded from this study. All the case children had received primary surgical treatment for oral clefts reconstruction during early childhood and have had free access since then to all medical teams including experienced pediatric, surgical, pediatric dentist, orthodontic, speech therapy and psychological support for children and their families.

The control group consisted of 35 healthy children, recruited at a Dental Clinic in Rio de Janeiro, Brazil. The control group was selected to ensure it had close similarity to the cleft group in terms of the number of children, age, gender and socio-economic status.

Ethical approval was obtained from Local Ethics Committee. Informed consent was obtained from all participating individuals or parents/legal guardians. The AUQUEI, quality of life assessment questionnaire, was applied as an interview, in the Portuguese language, while the children were waiting for their ambulatory follow-up visits. The questionnaire consists of 26 questions applied to the child himself/herself and assesses his/her satisfaction in different life circumstances, using 4 domains (autonomy, leisure, functions and family). The children were interviewed by a health care professional (F.M.M.S.), who was not a member of the cleft team. The children were asked to select the figure (very happy or happy or unhappy or very unhappy) that most corresponded to their feelings regarding a particular domain.

Parents or caregivers/guardians were asked what type of cleft their child had and their medical records confirmed the diagnosis (CL, CLP or CP). The socio-economic background of the population of the samples was summarized in terms of family income, using the minimum wage as a unit.

Data were analyzed using the Statistical Package for the Social Sciences (SPSS – 16.0). To compare the mean value of the AUQUEI scores of the two groups, we applied the T-test, and for comparative analyses of more than two groups we applied the ANOVA test. The

Table 1. Characteristics of the study children

	OFC children (n=35)	Non-cleft children (n=35)
Mean Age (SD)	8.34 (2.2)	8.37 (2.3)
Gender (%)		
Female	14 (40.0)	14 (40.0)
Male	21 (60.0)	21 (60.0)
Mean income of families (SD)	2.00 (1.0)	2.6 (1.8)
Type of cleft (%)		
Cleft lip (CL)	8 (11.4)	-
Cleft palate (CP)	7 (10.0)	-
Cleft lip and palate (CLP)	20 (28.6)	-
Quality of life (%)		
Good	26 (74.3)	22 (62.9)
Bad	9 (25.7)	13 (37.1)
AUQUEI score		
Minimum	39	37
Maximum	72	64
Mean (SD)	51.2 (7.3)	51.2 (6.8)

Chi-square test and Fisher exact test were used to analyze categorical variables. The level of statistical significance was set at $p \leq 0.05$.

RESULTS

The characteristics of the two groups are summarized in Table 1. The monthly family income of the majority of the children varied from less than 1 to 3 times the minimum wage, equivalent to USD 210–630.

The cleft group was made up of eight children with CL, 7 children with CP and 20 children with CLP. 25.7% of the OFC group experienced a low QoL and in the control group 37.1% experienced a low QoL. Within the cleft group, the CLP group reported a low quality of life for 35.0%, CP 14.3% and CL 12.5% ($p=0.3$). Figure 1 shows the mean scores for each combination of cleft type and age.

However the great majority in both groups reported a high quality of life. Quality of life was not positively associated with oral clefts in children ($p=0.44$). The mean score of AUQUEI in both groups was very similar ($p=1.00$) and is presented in Table 1.

There was no difference between cleft type mean scores ($p=0.61$).

There were no significant differences between the genders for the QoL in the OFC children and in the control group (Table 2). Table 3 shows the results of the four domains of the questionnaire related to the type of cleft.

DISCUSSION

This study provides data on the QoL of children with previously repaired OFC in comparison with controls without cleft. QoL in cleft children does not seem to be as bad as one would expect for the disease limitations. To our knowledge, this is the first study that uses an instrument to measure quality of life in Brazilian children previously treated for oral clefts. The reliability and validity of this instrument for children from 4 to 12 years old has been demonstrated in the literature,⁷ and is considered to be a valuable tool to

Table 2. Quality of life in both groups of children with gender controlled

Groups			Gender			P-value
OFC children	Quality of life	good	female	male	total	0.26*
		bad	12	14	26	
	total		2	7	9	
			14	21	35	
Non-cleft children	Quality of life	good	female	male	total	1.00*
		bad	9	13	22	
	total		5	8	13	
			14	21	35	

Note: * Fisher exact test

better understand the child’s experience in various aspects of his/her life, and for evaluation and follow up.⁶ Visual analogue scales are simple and easy to use, and present a continuous variable which is easily analyzed by researchers. They are often used to quantify the subjective assessment of global health or specific aspects of health such as pain.¹⁷

It is worth mentioning that the groups were homogeneous in terms of sample size, gender, age and socio-economic status. Our results reveal that both groups were similar in terms of quality of life. We observed that the majority children with clefts presented a high quality of life. We believe that this occurred because the cleft had been surgically repaired during earlier childhood and all children were included in a health care program in a reference hospital, with follow-up visits and psychological support to both the child and the families. Furthermore, comprehending the concept of quality of life is a complex task for children and even for adults⁹ which may explain the findings of the present study.

Locker *et al*¹² assessed the health related quality of life (HRQoL) of 11- to 14- year-old subjects with orofacial conditions. There were few differences in quality of life of subjects with orofacial conditions, compared with subjects with common dental conditions. The authors reported that the majority of these populations are well adjusted and able to cope with any adversity they experience as a result of their condition. In agreement, according to Topolski *et al.*,¹³ adolescents with facial differences confront significant challenges to their own self-identity while experiencing higher QoL from relationships, possibly from their need to negotiate and maintain close family support. Following these pattern, another instrument aims to create a craniofacial-specific module. Six of the domains (coping, stigma and isolation, intimacy and trust, positive consequences, self-image, and negative emotions) comprised the Youth Quality

of Life Instrument-Facial Differences module. One other domain, surgery, was a salient issue for many of the youth, but not all, so it was made into a separate module, the Youth Quality of Life Instrument-Craniofacial Surgery module.¹⁸ It is important to emphasize that our studied population included only cleft and non-cleft children (5-12 years) and these studies, cited above, reported quality of life of adolescents.

Our findings were not in accordance with a previous study¹⁶ which analyzed the quality of life in cleft children using the KINDL questionnaire. This questionnaire is a self-reporting instrument specifically developed to assess the age-related QoL in children. Several dimensions of QoL, such as physical wellbeing, psychological well being, self-esteem, family, friends and everyday activities were included. In this previous study, the authors observed that the quality of life of cleft children was superior to the control group. Boys with oral clefts were shown to experience a lower QoL when compared with girls. According to Hurrelmann,¹⁹ boys are less willing than girls to reveal their problems to others and are also less willing to ask for help, increasing the psychological distress. We did not observe a significant difference in the quality of life between the cleft and control group and we did not find differences between genders. The exact reason why gender differences in QoL were not seen in our study cannot be explained but, probably, the low age of the children included in this study can be influenced our results.

An earlier study reported that both in the preschool children’s KINDL and in the parents KINDL questionnaires, the lowest scores were observed in the dimension for self esteem.¹⁵ Another study, cited above, reported that reductions in QoL were observed in school age children with cleft lip and palate in the dimensions “family” and “friends”, indicating problems in the social field.¹⁶ Regarding the four domains analyzed here, we did not observe any differences

Table 3. Quality of life regarding of the four dimensions related to the type of cleft.

Dimensions	Non-cleft children		All Clefts		P-value	Type of Cleft					
	Mean	SD	Mean	SD		CL		CP		CLP	
Functions	9.9	2.0	10.3	2.0	0.81	10.8	1.2	7.6	2.5	10.3	1.5
Family	9.8	2.2	10.6	2.3	0.83	10.5	1.5	9.3	2.3	9.7	2.4
Leisure	7.1	1.4	7.7	1.2	0.40	6.7	0.7	7.1	1.7	7.2	1.5
Autonomy	7.9	2.4	6.9	1.8	0.26	8.5	1.0	7.4	2.6	7.9	2.7

Note: t-test was used and p-value presents the difference between “non-clefts” and “all clefts” children.

between the groups. Probably the limited sample size could have influenced our results. We did not find in the literature similar data to compare the QoL using AUQUEI in cleft children.

Our results demonstrate that cleft types did not differ in QoL. Considering only the frequencies related, CLP children reported a worse quality of life than CP and CL group. This finding could be explained by the severe speech disorders and facial esthetical problems in this group. And indeed speech and esthetic concerns seem to have been important factors affecting the health-related quality of life for children with oral clefts.¹⁴ The relationship between cleft type and QoL was more complicated, and varied according to a child's age group. The variations of scores in QoL for the cleft groups reflected the influence of age when physical appearance becomes more important for social interaction, in children with CL, and the improvement in speech is critical in children with CP.

Nevertheless, our case-control study has several limitations. First, our study was limited to the questionnaire used. The AUQUEI is a generalized QoL measurement without any oral health QoL components. Probably, it would be better to assess more typical QoL dimensions, such as appearance, social acceptance and self-esteem. An oral health QoL questionnaire assesses the direct impact of oral conditions on related outcomes such as appearance, oral function and social interactions. Second, the sample size could be considered small. This finding of no difference across cleft types and QoL may reflect the small sample size; especially for CL and CP patients. On the other hand, we believe that the rigorous inclusion criteria minimized the bias in our study. The understanding of the impact of clefts on children's QoL is an important issue and future studies may also explore QoL on the basis of initial cleft severity, in addition to cleft type. Case control studies where inclusion criteria are defined on basis of severity reduce selection bias. Additionally, extend study to include adolescents.

In summary, to increase the successful outcome of medical treatment for cleft children it should not be defined solely in terms of repair of facial and/or palatal clefting but should also be extend to the maintenance and improvement of the QoL of the patients and their families. In our work, children treated for cleft did not relate a low QoL, which suggests that health care programs associated with familial psychological support, are beneficial for this population.

Acknowledgments

The authors are indebted to the participants of the study. Support for this work was provided by CAPES (PNT and ECK, fellowship), by PIBIC/UFRRJ (FMMS fellowship), FAPERJ (MCC), FINEP, and CNPq (JMG).

REFERENCES

1. Murray JC. Gene/environment causes of cleft lip and/or palate. *Clin Genet*; 61:248-56. 2002.
2. Wyszynski DF, Wu T. Prenatal and perinatal factors associated with isolated oral clefting. *Cleft Palate Craniofac J*; 39:370-5. 2002
3. Habel A, Sell D, Mars M. Management of cleft lip and palate. *Arch Dis Child*; 74:360-6. 1996.
4. Broder HL, Smith FB, Strauss RP. Habilitation of patients with clefts: parent and child ratings of satisfaction with appearance and speech. *Cleft Palate Craniofac J*; 29:262-7. 1992.
5. Yazdy MM, Honein MA, Rasmussen SA, Frias JL. Priorities for future public health research in orofacial clefts. *Cleft Palate Craniofac J*; 44:351-7. 2007.
6. Manificat S, Dazord A, Cochat P, Nicolas J. [Evaluation of the quality of life in pediatrics: how to collect the point of view of children]. *Arch Pediatr*; 4:1238-46. 1997.
7. Assumpcao FB, Jr, Kuczynski E, Sprovieri MH, Aranha EM. [Quality of life evaluation scale (AUQUEI--Autoquestionnaire Qualite de Vie Infant Image). Validity and reliability of a quality of life scale for children 4 to 12 years-old]. *Arq Neuropsiquiatr*; 58:119-27. 2000.
8. Manificat S, Dazord A, Cochat P, Morin D, Plainguet F, Debray D. Quality of life of children and adolescents after kidney or liver transplantation: child, parents and caregiver's point of view. *Pediatr Transplant*; 7:228-35. 2003.
9. Neiva PD, Kirkwood RN, Godinho R. Orientation and position of head posture, scapula and thoracic spine in mouth-breathing children. *Int J Pediatr Otorhinolaryngol*; 73:227-36. 2009.
10. Castelo PM, Barbosa TS, Gaviao MB. Quality of life evaluation of children with sleep bruxism. *BMC Oral Health*; 10:16. 2010.
11. de Oliveira CM, Araujo AP. Self-reported quality of life has no correlation with functional status in children and adolescents with spinal muscular atrophy. *Eur J Paediatr Neurol*; 15:36-9. 2011.
12. Locker D, Jokovic A, Tompson B. Health-related quality of life of children aged 11 to 14 years with orofacial conditions. *Cleft Palate Craniofac J*; 42:260-6. 2005.
13. Topolski TD, Edwards TC, Patrick DL. Quality of life: how do adolescents with facial differences compare with other adolescents? *Cleft Palate Craniofac J*; 42:25-32. 2005.
14. Damiano PC, Tyler MC, Romitti PA, Momany ET, Jones MP, Canady JW, et al. Health-related quality of life among preadolescent children with oral clefts: the mother's perspective. *Pediatrics*; 120:e283-90. 2007..
15. Kramer FJ, Gruber R, Fialka F, Sinikovic B, Schliephake H. Quality of life and family functioning in children with nonsyndromic orofacial clefts at preschool ages. *J Craniofac Surg*; 19:580-7. 2008.
16. Kramer FJ, Gruber R, Fialka F, Sinikovic B, Hahn W, Schliephake H. Quality of life in school-age children with orofacial clefts and their families. *J Craniofac Surg*; 20:2061-6. 2009.
17. Harrison MJ, Boonen A, Tugwell P, Symmons DP. Same question, different answers: a comparison of global health assessments using visual analogue scales. *Qual Life Res* 2009.
18. Edwards TC, Patrick DL, Topolski TD, Aspinall CL, Mouradian WE, Speltz ML. Approaches to craniofacial-specific quality of life assessment in adolescents. *Cleft Palate Craniofac J*; 42:19-24. 2005.
19. Hurrelmann K. Health promotion for adolescents: preventive and corrective strategies against problem behavior. *J Adolesc*; 13:231-50. 1990.