

Development and Validation of the Quality-of-Life Adolescent Cleft Questionnaire in Patients With Cleft Lip and Palate

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Abstract: Only a few reports in the literature have described the use of specific instruments for assessing the quality of life in adolescents and young adults with cleft lip and palate (CLP). This condition markedly affects their lifestyle, even after surgical treatment. In the present study, we aimed to develop a quality-of-life assessment tool specifically designed for such patients with CLP. Our multidisciplinary team created a questionnaire focused on the physical, psychological, and social satisfaction of adolescents and young adults with CLP, which was adapted from 3 dimensions of the 36-item Short-Form Health Survey. The questionnaire was administered to a randomized sample of 40 adolescents and young adults (aged 16–24 years) with CLP who had completed treatment protocols and 40 (aged 16–24 years) who were not affected by CLP.

The statistical results stated that the questionnaire had good reliability and validity; the Cronbach α coefficient was found to be 0.944. Moreover, factorial analysis confirmed the presence of 3 subscales that were the fundamental components of this questionnaire, which is consistent with the areas theoretically proposed and from which the items were designed and selected.

Thus, we validated our novel questionnaire that was administered in the present study and proved its consistency. However, further investigations on a larger population would be useful to confirm these findings.

Key Words: cleft, quality of life, adolescent, questionnaire

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Only few studies have assessed the influence of cleft lip and palate (CLP) on learning, adjustment, and behavior. These studies have not directly assessed the simultaneous influence of facial disfigurement and speech difficulties on specific adjustment indices such as self-perception, anxiety, depression, and behavior.¹

Despite surgical advances and the advantage of undergoing surgical repair at a younger age, these patients are often affected by psychological issues due to their condition, particularly during adolescence.³ Although psychological consultation is available in many centers, few teams investigate the psychological issues that CLP patients experience.

In particular, self-perception and social skills are important for their psychological health.⁴ Self-esteem can be influenced by the manner in which people interact with individuals with CLP, who often experience social ostracism.⁵ Individuals with a persistently low self-esteem experience difficulties in participating in social activities and are considered as less intelligent or less socially acceptable.^{6,7}

Therefore, CLP patients should be treated by a multidisciplinary team comprising maxillofacial surgeons, plastic surgeons, speech therapists, dentists, geneticists, and psychologists. The main goal of multidisciplinary treatment is to achieve the best possible outcome for the patient and the family.¹

Several reviews have described different clinical instruments available for the assessment of the quality of life (QoL) of CLP patients,^{1,2} but only few are specific for adolescents, and none have been adopted and used in Italy.

In the present study, we aimed to develop, translate into Italian, and validate an instrument—the Quality of Life Adolescent Cleft (QoLAdoCleft) Questionnaire—that is specific for CLP patients who are adolescents or young adults.

METHODS

Study Design

A team of professionals (5 surgeons, 2 speech therapists, and 2 statisticians) studied the domains of the 36-item Short-Form Health Survey and developed the content of the questionnaire used in the present study.⁸ The content is divided into 2 sections: one containing the clinical profile (anamnesic data: first/second name, age, sex, education level, parents' educational level, cleft type, and surgery performed) and the second focusing on physical, psychological, and social health. Each area consists of subsets, and every subset was used

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The authors report no conflicts of interest.

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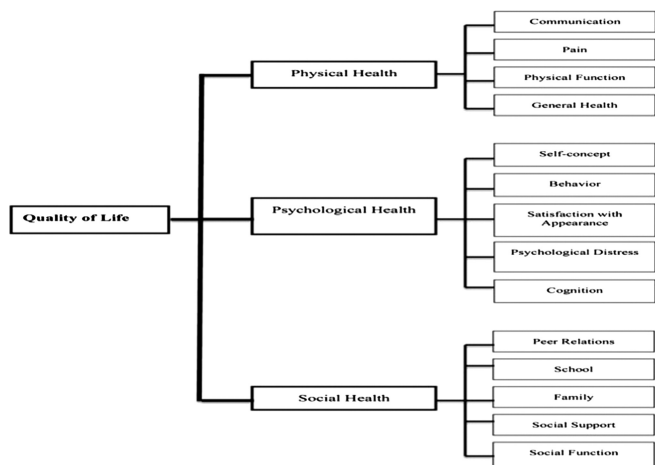


FIGURE 1. Structure of the QoLAdoCleft Questionnaire.

to evaluate the patient’s condition before and after adult orthopedic-orthodontic treatment, although the questionnaire was administered only after surgery in all the cases. Thus, our questionnaire was structured according to the proposal of Klassen et al (Fig. 1).² The answers were recorded using the Likert Scale (values from 0 to 4, with 0 representing the best attitude possible and 4 representing the worst).⁹

Study subjects were interviewed by telephone by 1 researcher in each center. A parent was present during the interview for all study participants younger than 18 years. The selected cluster was first administered the QoLAdoCleft Questionnaire and then retested with the Child Oral Health Impact Profile (COHIP)^{8bis} Questionnaire on the same day in order to assess the reliability of our test. A blind statistical analysis was performed on the raw data.

Moreover, a literature review was conducted by searching MEDLINE, PubMed, Cochrane Database, Elsevier Science Direct, Ovid, Wiley, and JSTOR for the keywords “quality of life and cleft,” “quality of life questionnaire cleft,” “QoL adolescents cleft,” and “QoL cleft lip and palate.”

Participants

The questionnaire was administered in 3 different cleft centers (Naples, Bologna, and Rome). The study population included white adolescent and young adult patients aged from 16 to 24 years who had completed surgical treatment and had a similar clinical profile. The sample was extracted from a list of 106 cases from Naples, Bologna, and Rome, by using a simple random sampling scheme. We included, in the list, patients with a clinical diagnosis of CLP, cleft lip, or cleft palate; patients aged between 16 and 24 years at the end of treatment; and patients who completed orthopedic-orthodontic treatment but were not managed by a multidisciplinary team. Moreover, the patients’ parents were alive and not divorced. We excluded patients with previous physical trauma, patients with 1 or both parents deceased, patients with current legal problems or those having parents with current legal problems, patients whose cleft was associated with a clinical syndrome, patients with other coexistent congenital anomalies, and patients with previously diagnosed psychological disorders. We extracted from the numerosity study 50 patients: 6 patients refused to be included in the study, 4 were not reachable by phone (number change), and 40 patients agreed to answer the questionnaire. So, the sample size was 40, and the confidence level was 96%.

The sample included 19 males and 21 females (mean age, 20 years) who had completed surgical treatment and had a similar clinical profile: 19 patients with CLP (13 males, 6 females), 10 patients with cleft lip (3 males, 7 females), and 11 patients with cleft palate (3 males, 8 females).

The control group comprised 40 adolescents (19 males, 21 females; mean age, 20 years; range, 16–24 years). Control subjects for these cases have been appropriately drawn from the population of the same area in the same sex and age groups. They were patients under treatment or already treated for dental malocclusion at the Federico II University Hospital of Naples, S. Orsola-Malpighi Hospital–University of Bologna, and Santo Spirito Hospital, Rome. The catchment area was in the same geographical area of study group. This method allowed us to select a control group as weighted as possible with the study group: picking patients’ peers, meant comparing 2 individuals living the same social context and having a similar status quo, except for the facial abnormality.

Written informed consent was obtained from each patient, and the study was conducted in accordance with the Declaration of Helsinki. The study features were approved by a council of senior specialists at each cleft center.

Statistical Methods

The questionnaire was administered to a sample of 40 patients with CLP. Cronbach α was used to test the internal consistency and reliability (values of >0.7 were considered acceptable).¹⁰ The subjects were then administered the COHIP. The *P* values were estimated to identify whether significant differences were present between the results of the 2 questionnaires.

The control group was also administered the questionnaire. For each macro area and for each subset, the mean and the median values were calculated and illustrated on graphs using a box plot system. To validate the questionnaire, the statistical team used the bootstrap estimates. All the analyses were performed using a bootstrap sample of 1000 units. The bootstrap confidence interval is calculated by simply taking bootstrap samples with replacement from data, while calculating for each α , and computing the quantiles.

Reliability was determined according to the internal consistency. The questionnaire’s global scale was found to have high internal consistency. The reliability of the subscales was tested in the same manner. In all the subscales, all reproducibility modes showed a consistently high correlation index of more than 0.90; the internal consistency was also found to be high.

In the assessment of validity, the presence of 3 subscales that were the fundamental components of this questionnaire was identified by factorial analysis; this is consistent with the areas theoretically proposed earlier, from which the items were designed and selected.

RESULTS

All the data were analyzed by adding scores obtained from each macro area and for each subset. The Cronbach α value (for internal consistency and reliability) was calculated, and this value for the QoLAdoCleft Questionnaire was 0.941.

The mean and median values of the scores obtained for each macro area were calculated and are provided in Table 1. The mean and median values were also estimated for each subset because they are the main measures of central tendency of a distribution and thus enabled comparisons of the data. The same data were assessed after division into 2 subsets: before and after orthopedic/orthodontic treatment (Fig. 2).

TABLE 1. Rough Data From the Macro Areas of the QoLAdoCleft Questionnaire

	Mean	Median
Average value of “physical health” score	2.469	2.267
Average value of “psychological health” score	3.013	2.644
Average value of “social health” score	2.536	2.675

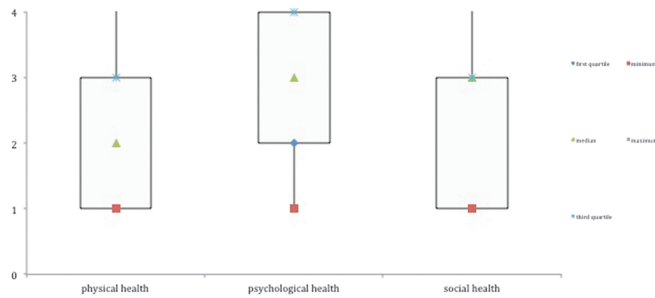


FIGURE 2. The 3 areas of the questionnaire.

Physical Health

Oral function difficulties were observed in 22.5% of patients before treatment, which decreased to 2.4% after treatment. Food reflux was noted in 30% of patients before treatment, but only 5% after treatment. For the subset “language,” a median of 3.65% of the samples showed frequent or persistent difficulties in articulating words and frequent misunderstanding by interlocutors (before treatment, 57.5%; after treatment, 10%). Timbre was perceived as nasal in 17.5% of patients before treatment, which decreased to 2.5% after treatment. We noted that 80% of patients did not report pain before treatment; this value increased to 90% after treatment. The subset “general health” showed a median value of 2 according to the semantic scale from 0 to 4 used in the questionnaire—the value of 2 corresponded to “adequate general health.”

Psychological Health

The domain “psychological health” showed a median value of 3. In the subset “self-confidence,” 12.5% of participants felt less socially acceptable as compared with healthy people, before and after treatment. Of those interviewed, 5% did not feel different from their peers before treatment; this value increased to 10% after treatment.

In the “behaviors” subset, a tendency for isolation (before treatment, 57.5% of subjects tended to be isolated indifferent social contexts) and discomfort (only 27.5% said they never experienced discomfort in relationships with each other) was reported.

In the subset “psychological stress,” anxiety (27.5% of adolescents are often anxious about their state of health), depression (25% have “sometimes” manifested depressive symptoms, 15% manifest them “often,” and 5% “always” manifest such symptoms), and concern were assessed. However, 72.5% of respondents stated that they had never received psychological therapy support during the rehabilitation process.

The “appearance” category showed a median value of 4 and had the highest number of responses; the subjects were minimally (45%) or not at all (15%) satisfied with their facial appearance. With

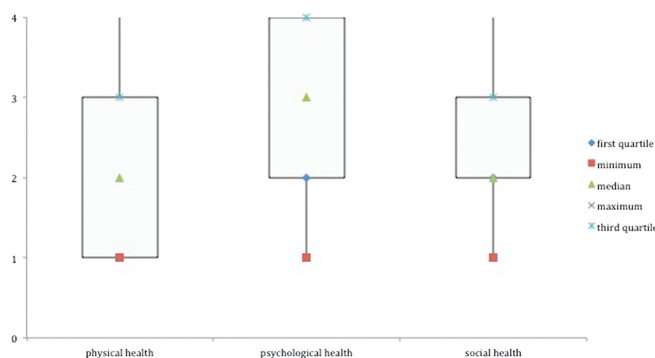


FIGURE 3. The physical, psychological, and social health subsets of the COHIP.

regard to oral cavity appearance, 27.5% were not satisfied; only 17.5% of the patients indicated that they did not need further treatment.

Social Health

The median value of 3 in the first subset indicates that 42.4% of patients were bullied, and 37.5% of patients were not satisfied with his/her life. Almost half (47.5%) of the patients felt that they will never be satisfied with their life. With regard to results in the box plot for “family function” concerning cleft management, we noted that 20% had family conflicts, 40% felt that their relationship with close relatives had changed because of the treatment, and 10% were unsatisfied with their relationship with their family members.

In the ‘Social Support’ subset, 52.5% of participants reported that they were supported by other people and institutions, whereas 42.5% of participants felt that they always received the help they needed from family, peers, and colleagues.

In the subset “peer relationships,” 30% of patients stated that could not relate to their peers, whereas 47.5% could not relate to their superiors.

The last box plot “structures” has median value of 3. Of the patients with difficulties in accessing specialized centers for cleft care, 32.5% did not go through an adequate care process, and 35% did not receive sufficient information on pathology management.

COHIP Questionnaire

Patients were also administered the COHIP Questionnaire (Cronbach $\alpha = 0.829$). The items common between the COHIP and QoLAdoCleft Questionnaires were compared. The *P* value obtained was 0.052 ($P > \alpha$), and therefore, the tests can be considered to be similar (Fig. 3).

Tests on Adolescents Without CLP

After the COHIP Questionnaire was administered, 40 controls were administered the QoLAdoCleft Questionnaire. The domain “physical health” showed a median value of 0. The domain “psychological health” showed a median value of 0.

The subset “aspect” showed that 12.5% of adolescents felt that they needed corrective surgery. In the domain “social health,” 10% of adolescents stated that they were bullied, 12.5% “sometimes” had difficulties in relationships with peers, and 20% had difficult relationships with superiors.

The results obtained from patients and controls were compared. The Likert scale scores ranged from 2 to 3 for patients and 1 to 2 for control subjects.

Reliability (Statistical Appendix)

The Cronbach α coefficient—both nonstandardized and standardized—was calculated for the global scale, for each of the subscales and for each of the items. The α coefficient value was 0.944 (standardized, 0.947) for the global scale, 0.876 (standardized, 0.880) for the physical health subscale, 0.897 (standardized, 0.898) for the psychological health subscale, and 0.903 (standardized, 0.907) for the social health subscale (Table 2).

TABLE 2. Reliability Statistics: Cronbach α on Subsets of Questionnaire

	Cronbach α	Cronbach α Based on Standardized Items	No. Items
General	0.944	0.947	50
Physical health	0.876	0.88	19
Psychological health	0.897	0.898	14
Social health	0.903	0.907	17

The values of the α coefficient for the global scale, when each item was omitted from the scale, varied between 0.941 and 0.946. None of the items, when removed, caused an appreciable modification of the global internal consistency.

Validity (Statistical Appendix)

Factorial analysis confirmed the presence of 3 factors, which together explained 50% of total variability (first factor, 31%; second factor, 12%; third factor, 6% [Table 3]). The first factor was associated with items on the physical health subscale, the second factor was associated with items on the psychological health subscale, and the third factor was associated with items on the social health subscale. This finding is confirmed by the scree plot (Fig. 4). Considering the estimated factor model, the commonality indicated the share of explained variance of each indicator. The values of the communalities for the global scale varied between 0.733 and 0.924. All the variables were represented very well in the factor solution that considered the 3 main components.

DISCUSSION

This epidemiological survey indicated the importance of certain variables that negatively affect the QoL of patients with CLP. These variables have been assessed and examined by previous studies.^{1,2,11}

In the domain “physical health,” the subjects reported low scores (a good self-perception); these scores increased for the subset “language.” Millard and Richman^{1,12} noted that speech and facial appearance were related to the overall adjustment of children with CLP.^{1,12} However, patients examined by Millard were aged 7 to 18 years, whereas we examined patients at a critical age (ie, adolescence; 16–24 years); moreover, the results of patient self-esteem obtained in our study was worse compared with that obtained in the study by Millard.

The degree of speech difficulty in children with CLP may contribute to their low self-esteem, self-perceived depressive symptoms, and anxiety.¹ We noted that CLP patients were more likely to self-report higher scores (such as for bad attitude) on their pathology perception, which is consistent with previous studies.¹³ The domain “psychological health” showed higher values (such as for bad self-perception). As stated by Turner et al,⁴ patients with CLP have problems in relating to their peers, because they are perceived as being different. According to Ramstad¹⁴, Bloss¹⁵, and Brother¹⁶, this finding is due to a negative self-perception (ie, they feel less socially acceptable than their peers). The physical appearance appeared to be a major problem, according to Bernstein et al.¹⁷ This finding is not consistent with the outcome of Millard’s study,¹ presumably due to the difference in patient age between the studies. According to Bernstein and Kapp^{17,18} and Belfer et al,¹⁸ these patients will require additional surgical treatment to improve their physical appearance. Physical problems appear to positively correlate with social difficulties;¹⁹ the present study confirmed that CLP patients are bullied more frequently than their peers, which has also been reported by Hunt et al.²⁰ However, encouraging scores were noted from the subset “family”; CLP patients seemed to be satisfied with their family relationships, consistent with that noted in previous studies.¹⁶

TABLE 3. Validity—Cumulative Variance

Component	% of Variance	Cumulative %
1	31.264	31.264
2	12.575	43.839
3	5.956	49.794

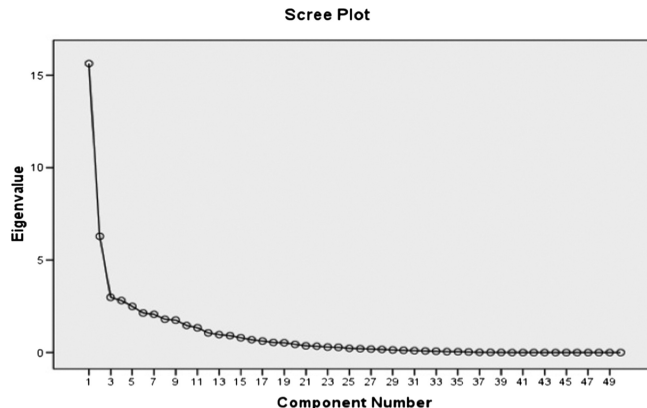


FIGURE 4. Scree plot.

A comparison between scores obtained in the study and the control group indicated that patients with CLP have more problems with self-esteem and social skills, consistent with previous studies in the literature.^{7,12,20–24}

However, the most interesting finding of the present study is that patients with CLP report having difficulty accessing information and specialized centers about their pathology. These findings require further study by other cleft centers in Italy.

To our knowledge, the present study was the first to evaluate the QoL of adolescent and young adults with CLP in Italy, by using a specific questionnaire specifically designed for them. The questionnaire was useful, easy to administer, and easy to interpret. However, because this study involved only 3 centers in Italy, the findings should be carefully interpreted. Therefore, additional studies in other centers are required, particularly studies evaluating patients treated by multidisciplinary teams.

CONCLUSIONS

We developed an instrument—the QoLadoCleft Questionnaire—for determining the decline in QOL in patients with CLP who are not treated by multidisciplinary teams. We aimed to elucidate the manner in which a multidisciplinary team approach in CLP can be made mandatory and the criterion standard for treatment to achieve optimal outcomes. However, further research is required to assess the needs of patients with CLP and to help medical care professionals improve the outcome of the treatment provided. Furthermore, this questionnaire should be validated for use in other centers as well.

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