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Original article

Development of a valid and reliable instrument for the assessment of quality of life in parents of children with clefts

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ABSTRACT

Purpose

Orofacial clefts are the most common congenital malformations that affect craniofacial structures. Studies show that they have a major influence on psychological development of the patient, and on their families. A review of the literature showed a lack of specific questionnaires for children and their parents. This study investigated the impact of orofacial clefts in children on the quality of life of their parents. In addition, the results of the treatment and the quality of work of the health team members involved in this process were evaluated.

Materials and methods

For the purpose of this study, an original questionnaire was made to analyse the effect of orofacial clefts in children who had undergone surgery on the quality of life of 73 of their parents. The questionnaire consisted of 28 simple statements, which were evaluated with a 5-degree Likert scale (from 1–fully disagree to 5–fully agree), did not require any specific additional clarification, and were easy to complete.

Results

Analysis of areas of the questionnaire that applied to the parents, resulted in two subscales, parental social health and child social health, which had satisfactory Cronbach's coefficients (0.907 and 0.897, respectively). However, some issues had a relatively poor coefficient of internal consistency, which justified their expulsion from the final model of the parent questionnaire.

Conclusion

The questionnaire developed for this study comprised two subscales concerned with the social health of parents/respondents and the social health of adolescents, as perceived by the parents. It was a valid and reliable instrument, and it showed satisfactory quality of life for parents of adolescents with clefts.

1. Introduction

Orofacial clefts are the most common congenital malformations that affect craniofacial structures. Failure of tissue fusion during embryological development results in aesthetic and functional problems. Clefts can be a result of a large number of syndromes, but can also be non-syndromic, i.e., isolated events. For both these scenarios, cases are divided into cleft lip, cleft lip and palate or isolated cleft palate [1]. Children born with these types of malformation have difficulties with talking, hearing, dental irregularities and characteristic changes in the appearance of their nose and upper lip, affecting all domains of health. Studies [2–5] show that orofacial clefts have a major influence on psychological development of the patient, and on their families. The treatment of children with clefts involves a large number of specialists whose activities are intertwined as the child grows up, including a maxillofacial surgeon who coordinates other team members, a neonatologist, an anaesthesiologist, a paediatrician, an orthodontist, an oral surgeon, an otorhinolaryngologist-audiologist, a logopedist, a psychologist, a psychiatrist, and a paediatric dentist [6].

According to the latest data, the incidence of orofacial clefts is about 1 in 700 children born in the world [7]. In Croatia, according to a study by Magdalenić-Meštrović's and Bagatin in 2005, the incidence is somewhat higher, at 1 in 581 births [8]. Cleft lip, with or without cleft palate, is more common in males, while isolated cleft palates are more often found in females, regardless of ethnicity. In Caucasians, cleft lip, with or without cleft palate, is more common in males by a ratio of 2:1 [7].

In 1948, the World Health Organization defined health as a state of complete physical, mental and social well-being, not just the absence of disease and infirmity [9]. American psychologist John C.

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Flanagan is considered to be the first to develop the concept of quality of life in health care [10]. Health related quality of life (HRQOL) describes a subjective satisfaction with one's health status. It is an instrument that examines the impact of illness and treatment modalities on health, integrating an objective assessment of health status with its subjective experience (physical, mental and social functioning) [11].

The concept of oral health-related quality of life (OHRQOL) has only been developed over the past few decades. Oral health is considered an important part of the general health of a patient [12].

A review of the literature shows that there is a large number of quality of life studies targeted at people of different ages with clefts, as well as their parents, [13] and also, a large number of generic questionnaires used as metrics. However, there is a lack of specific questionnaires for children with clefts, and their parents. In Croatia, only Vuletić et al have conducted a study on this issue [14]. The current study investigated the impact of orofacial clefts on the quality of life of parents of children with clefts. In addition, it evaluated the results of the treatment and the quality of work of the health team members involved in this process.

2. Materials and methods

Ethical approval for this study was obtained from the Ethics Committee of the University Hospital Dubrava, Zagreb, Croatia.

2.1. Questionnaire construction

For the purpose of this study, an original questionnaire was made to analyse the quality of life of parents of children who had undergone surgery for orofacial clefts. It was constructed in cooperation with parents of children with clefts, by a maxillofacial surgeon, an orthodontist, a psychologist and a logopedist, who have long-term experience in treating clefts. Many generic questionnaires that examined the quality of life of parents/guardians were studied, such as the Impact on Family Scale (IOFS), [15] and the Child Behavior Checklist (CBCL) [16].

Initially, the literature was reviewed through PubMed and Medline for keywords, such as “quality of life and clefts”, “quality of life family/caregivers”, “quality of life questionnaires cleft”, from January to March 2017. Questions were then created concerning quality of life of parents of adolescents with clefts. Initially, about 50 questions were formulated. The validity of the questionnaire content was discussed by all members of the cleft team, and the final number of questions/statements was reduced to 28. The questions/statements were carefully designed and chosen by the study group to reflect the main goals of the research.

The questionnaire used in the current study was titled The Quality of Life of Parents of Adolescents with Cleft Assessment (QLPACA). It consisted of simple statements, which were evaluated with a 5-degree Likert scale (from 1–fully disagree to 5–fully agree). As such, they did not require any specific additional clarification, and were easy to complete.

2.2. Subjects

QLPACA was administered to 80 parents of children who underwent cleft surgery between June 2017 and June 2018, during follow-up examinations of their children at the Clinical Department for Oral Surgery and the Clinic of Maxillofacial Surgery at the Clinical Hospital Dubrava, Zagreb, Croatia. Written informed consent was signed by each parent/guardian that participated. Three parents refused to take part and four parents did not answer all the questions; therefore, they were excluded from the study.

Inclusion criteria for this study were that participants were parents of children with non-syndromic clefts that had undergone surgery, the children were without learning disorders or other medical problems and they showed continuity of medical treatment. Exclusion criteria for the study were that their children had a cleft that had not undergone surgery, was the result of a syndrome, or they had learning disabilities or other medical problems.

2.3. Statistical methods

Differences in categorical values were analysed by χ^2 test, while differences in quantitative values were analysed by independent t-test, after checking for normality with the Kolmogorov–Smirnov test. In order to reduce the number of variables, i.e., space dimensions, exploratory factor analysis was used with the main component method. The criterion for rejecting new variables that provided little information, or retaining those that provided most of the information contained in the variables, included: Kaiser's criteria for taking only those main components corresponding to eigenvalues greater than 1; and the Cattell screen plot, where the shape of the diagram was determined by the number of factors, the percentage of the explained variant according to which the factors are excreted as long as there are no longer determinable factors that significantly reduce the unexplained variation, the retaining of the components which carry or retain each particular predetermined part of the information (criterion meaning fullness or interpretability). Each of these criteria could be applied as a rule to reduce variables, and their choice in a particular case was determined by ensuring a non-excessive number of variables were retained, and that the loss of information from the starting system was as small as possible. All factors that met the principles of all three criteria are retained, and explained more than 55% of the variance. Factor saturation and distribution of individual parts of questionnaires were shown after applying a varimax rotation. Cronbach's α coefficient was used to measure internal consistency of single factors. It was calculated for each factor separately, with its total value also being determined, and only those domains having a value > 0.700 were included for further interpretation. Higher values of Cronbach's α coefficient indicated greater reliability. The validity of individual factors was determined by corrected inter-item correlation, with the interrelationship between the arithmetic means, the variances and the Cronbach α coefficient, which for certain factors were approximately the same. In order to evaluate the usable value of the questionnaire, all factor scales, including the total score scale, were converted to a range of 0–100, where 0 was no exposure/subjective perception of the specified stress factor, and 100 indicated the highest possible exposure/subjective perception. Pearson's correlation coefficients were calculated between the separated factors and for the total questionnaire. *P*-values below 0.05 were considered statistically significant. STATISTICS, Version 12.0 was used in the analysis Fig. 1.

3. Results

The gender and the age characteristics of the study group are shown in Table 1. In most cases (63.0%) the respondents were mothers, while slightly more than a half of them were aged between 41 and 50 years. The average scores for the whole group of respondents/parents are shown in Table 2. Critical questions were those with an average above 3, and these were highlighted (Table 2).

Cronbach's α coefficient for this questionnaire applied to parents, the influence of individual parts on the overall coefficient of internal

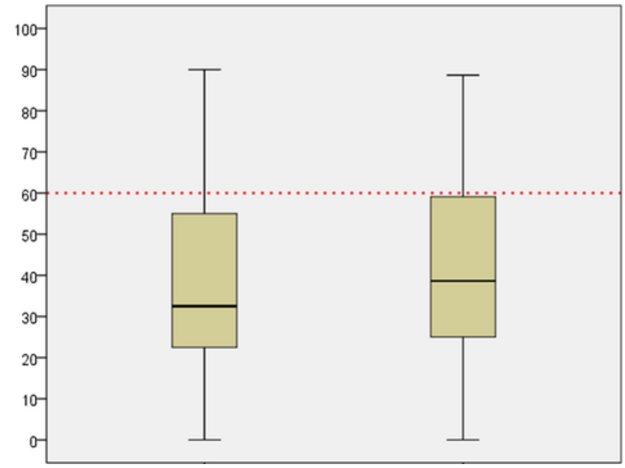

Fig. 1. Distribution of the scores for domains social health of child (left) and social health of parents (right), and the limit values that includes at least 75% of parents.

Table 1
Gender and age characteristics of parents of the examined group.

	N	%
Gender		
Male	27	37.0%
Female	46	63.0%
Age		
≤ 4 0 years	22	30.1%
41–50 years	39	53.4%
> 50 years	12	16.4%

consistency, and the summaries of the questionnaire for parents are presented in Tables 3 and 4. While the overall Cronbach coefficient was satisfactory (0.898), the following questions were excluded from the final questionnaire model, because they significantly reduced the Cronbach's coefficient:

- 25. My child does not agree with me and/or with other family members.
- 28. I did not provide the best medical treatment for my child that I could afford.
- 17. The approach of the doctors hindered my explanation of the difficulties my child is experiencing.
- 7. We have been weakened as a family because of this experience.
- 12. In caring for the condition, I do not look after myself.
- 15. The health status of my child is affecting my relationship with them.
- 13. I cannot imagine having more children because of my child's cleft.

These questions were probably confusing to parents due to their structure (negation), and their responses were inconsistent.

Assessment of particular categories of the questionnaire, obtained by factor analysis after varimax rotation, and the classification of coefficients by strength, is shown in Table 5. The questions were grouped in two categories, parental social health and child social health, with satisfactory Cronbach's coefficients of 0.907 and 0.897, respectively. However, some issues had a relatively poor internal consistency coefficients, which justified their expulsion from further analysis.

Apart from mutually significant correlations between individual domains, there was no significant correlation with the age of the respondents (Tables 6 and 7).

Table 2
Average response values obtained by parent questionnaire (response range of 1 = completely disagree, 5 = completely agree).

	N	Arithmetic mean	SD
1. I have to reduce my working time so that I can take care of a child with a cleft	73	2.90	1.48
2. It is more difficult to focus on my job because of the growing needs of my child	73	3.00	1.34
3. The cleft in my child causes financial problems in my family	73	3.04	1.37
4. My wife/husband and I have less time for ourselves due to our greater concern for our child	73	3.00	1.40
5. The cleft in my child is the reason we do not socialise enough with our friends	73	2.38	1.28
6. Family activities are reduced	73	2.40	1.15
7. We have been weakened as a family because of this experience	73	1.77	1.03
8. It is more difficult for my child to find friends because of their appearance	73	2.23	1.20
9. I have no time for other family members	73	1.95	1.01
10. I'm often exhausted and work-worn	73	2.73	1.30
11. I live day by day and I do not plan the future	73	2.38	1.27
12. In caring for the condition, I do not look after myself	73	2.71	1.27
13. I can not imagine having more children because of my child's cleft	73	1.92	1.24
14. I feel enormous pressure that nobody understands	73	2.82	1.43
15. The health status of my child is affecting my relationship with them	73	2.40	1.43
16. It is hard for me to take care of another child/children because I pay lots of attention to my child with a cleft	73	2.27	1.23
17. The approach of the doctors hindered my explanation of the difficulties my child is experiencing	73	1.71	0.92
18. People who know us do not forget about my child's appearance	73	2.47	1.30
19. It is difficult to find a person who is willing to take care of a child with a cleft	73	2.38	1.21
20. People sometimes do not understand my child because of their difficulty in pronouncing words	73	2.78	1.38
21. I think my child is not happy with her/his appearance	73	3.00	1.36
22. Other kids mock my child.	73	2.22	1.15
23. My child thinks everybody else is better than her/him	73	2.36	1.23
24. My child is too shy	73	2.60	1.26
25 My child does not agree with me and/or with other family members	73	2.56	1.29
26. I think my child will be less likely to find a girl/wife/boy/husband than her/his peers	73	1.97	1.27
27. I think my child is concerned about her/his future	73	2.49	1.32
28. I did not provide the best medical treatment for my child that I could afford	73	1.70	1.01

Table 3
Cronbach's α reliability coefficient for questionnaire applied to parents.

Cronbach's α coefficient	Number of parts
0.898	28

4. Discussion

In previous research, a valid and reliable instrument for the assessment of the quality of life of adolescents with cleft lip/palate was developed [14]. By contrast, the current study developed a reliable instrument for the assessment of quality of life among parents of adolescents with cleft lip/palate, which also evaluated their perception of the quality of life of their children.

Table 4
Influence of individual statements on total coefficient of internal consistency and compilation scale for parents questionnaire.

	The average value of the scale if the statement was dropped	The variance of the scale if the statement was thrown out	Corrected correlation of the statement with the scale	Cronbach's α coefficient if the statement was thrown out
1. I have to reduce my working time so that I can take care of a child with a cleft	65.25	306.33	0.486	0.895
2. It is more difficult to focus on my job because of the growing needs of my child	65.15	301.21	0.659	0.891
3. The cleft in my child causes financial problems in my family	65.11	306.46	0.531	0.894
4. My wife/husband and I have less time for ourselves due to our greater concern for our child	65.15	293.91	0.787	0.888
5. The cleft in my child is the reason we do not socialise enough with our friends	65.77	304.79	0.614	0.892
6. Family activities are reduced	65.75	308.22	0.599	0.893
7. We have been weakened as a family because of this experience	66.38	329.57	0.083	0.901
8. It is more difficult for my child to find friends because of their appearance	65.92	307.66	0.588	0.893
9. I have no time for other family members.	66.21	315.11	0.491	0.895
10. I'm often exhausted and work-worn	65.42	301.33	0.679	0.891
11. I live day by day and I do not plan the future	65.77	315.07	0.381	0.897
12. In caring for the condition, I do not look after myself	65.44	343.14	- 0.233	0.908
13. I can not imagine having more children because of my child's cleft	66.23	318.46	0.310	0.898
14. I feel enormous pressure that nobody understands	65.33	300.11	0.639	0.891
15. The health status of my child is affecting my relationship with them	65.75	331.05	0.013	0.905
16. It is hard for me to take care of another child/children because I pay lots of attention to my child with a cleft	65.88	305.30	0.628	0.892
17. The approach of the doctors hindered my explanation of the difficulties my child is experiencing	66.44	329.53	0.102	0.901
18. People who know us do not forget about my child's appearance	65.68	304.16	0.614	0.892
19. It is difficult to find a person who is willing to take care of a child with a cleft	65.77	306.07	0.620	0.892
20. People sometimes do not understand my child because of their difficulty in pronouncing words	65.37	304.90	0.561	0.893
21. I think my child is not happy with her/his appearance	65.15	312.24	0.408	0.896
22. Other kids mock my child.	65.93	308.06	0.607	0.893
23. My child thinks everybody else is better than her/him	65.79	302.42	0.698	0.890
24. My child is too shy	65.55	311.25	0.473	0.895
25 My child does not agree with me and/or with other family members	66.18	320.82	0.250	0.899
26. I think my child will be less likely to find a girl/wife/boy/husband than her/his peers	65.59	306.05	0.577	0.893
27.I think my child is concerned about her/his future	65.66	300.42	0.688	0.890
28. I did not provide the best medical treatment for my child that I could afford	66.45	328.95	0.104	0.901

The QLPACA questionnaire met the quality criteria for the evaluation of health status questionnaires [17]. Content validity and internal consistency were recognised as the most important properties of health status questionnaires [17]. The measurement aim and target population were clearly defined, and experts and parents of children with clefts were involved in item selection, ensuring the validity of the content.

Factor analyses were performed and Cronbach's α coefficients were between 0.70 and 0.95. Internal consistencies of seven answers were found to be relatively poor; therefore, the corresponding statements were excluded from further analysis. The internal consistency of the reduced questionnaire for the evaluation of quality of life was shown to be satisfactory.

The questionnaire demonstrated that the parents of children with cleft lip/palate had a satisfactory quality of life. Only four of the 28 statements had scores with an arithmetic mean > 3, indicating a reduced quality of life. Among these, the statement "Cleft causes financial problems in my family" scored the highest, which was interesting because in Croatia the complete treatment cost was covered by a national social health insurance fund, the Croatian Institute for Health Insurance, unlike in some developed countries where treatment of this type of malformation imposed a more personal financial demand [18–20]. It was possible that parents of children with clefts in wanting to give their child the best possible care, also sought assistance from private specialists who work outside the social health care system, creating additional costs. In addition, one of the parents could have been unemployed, or might have had to miss work for frequent follow-ups at various specialists. Notably, the statement “I think my child is not happy with her/his appearance” had a high score in parents, which was consistent with the opinion of their children in previous research [14]. Contrary to this knowledge, Hunt et al. indicate that children with clefts are more satisfied with their looks than their parents think, while parents of children without a cleft from the control group are more satisfied with the appearance of their children than that of themselves [21].

Factorial analysis of the questionnaire identified two areas/domains, the social health of the parent and the social health of the child. The overall Cronbach's coefficient was 0.898. Questions in the area of social health of parents were focused on examining the influence of orofacial clefts on the quality of life of parents, through relationships with other members of the family, social functioning, social support and personal functioning. The analysis showed that their social health was not disturbed by having to care for a child with an orofacial cleft. Their relationship with their family and the environment, as well as their psychological status, indicated a good quality of life. By contrast, previous research showed that the birth of a child with a cleft might have a number of negative effects on the quality of life of a parent, including shock, sadness and guilt, [22] sensitivity to environmental influences, [23] and concerns about further complications and overall treatment [24]. Children with orofacial clefts often have impaired social health, including their relationship with parents, which also exacerbates the quality life of their parents, causing psychological difficulties for one or both of them [3]. Unlike the aforementioned difficulties that reduce quality of life, Collett et al. found no difference between the response of parents of children with clefts and the control group of parents with unaffected children [25]. Correlation of areas of social health of parents with sociodemographic parameters of gender and age did not show significant differences. By contrast, Nidey et al. found gender differences [26]. Fathers had greater confidence and were less concerned about the social stigma, and mothers often sought help and talked to others in order to cope with the problems that occupied them. Apart from the fact that a cleft can have a direct impact on the life and health of parents, especially mothers, it is an important indicator of the psychological health of the child, [5] and the mental health of parents also has a direct impact on the child's health and their level of education [27,28].

The second area/domain focused on the social health of the child, and contained 10 questions. It analysed the effect of teasing by other children, satisfaction with their appearance, speech, attitude towards the opposite sex, and shyness in company. From a review of the literature, [29–31] it has been found that teasing due to appearance has a high incidence in children with clefts, representing a basis for psychosocial difficulties. However, this was not corroborated by results

Table 5
Overview of particular areas of the questionnaire applied to parents obtained by factor analysis after varimax rotation and classification of coefficients by strength.

	Areas		
	Social health of parents	Social health of child	Excluded questions
4. My wife/husband and I have less time for ourselves due to our greater concern for our child	0.786	0.388	0,072
3. The cleft in my child causes financial problems in my family	0.742	0.091	−0,065
16. It is hard for me to take care of another child/children because I pay lots of attention to my child with a cleft	0.741	0.156	0,154
2. It is more difficult to focus on my job because of the growing needs of my child	0.716	0.266	0,029
6. Family activities are reduced	0.715	0.227	−0,056
10. I’m often exhausted and work-worn	0.714	0.351	−0,079
1. I have to reduce my working time so that I can take care of a child with a cleft	0.711	0.096	−0,158
5 The cleft in my child is the reason why we do not socialise enough with our friends	0.706	0.209	0,196
9. I have no time for other family members	0.560	0.185	0,101
14. I feel enormous pressure that nobody understands	0.532	0.466	−0,013
11. I live day by day and I do not plan the future	0.492	0.183	−0,131
22. Other kids mock my child	0.146	0.779	0.063
23. My child thinks everybody else is better than her/him	0.327	0.755	0.038
20. People sometimes do not understand my child because of their difficulty in pronouncing words	0.177	0.709	0.078
24. My child is too shy	0,105	0.704	−0,010
8. It is more difficult for my child to find friends because of their appearance	0,229	0.703	0.045
18. People who know us do not forget about my child's appearance	0.289	0.691	0.067
26. I think my child will be less likely to find a girl/wife/boy/husband than her/his peers	0.345	0.626	−0.203
21. I think my child is not happy with her/his appearance	0.082	0.600	−0.048
19. It is difficult to find a person who is willing to take care of a child with a cleft	0.361	0.567	0.129
27. I think my child is concerned about her/his future	0.531	0.555	−0.114
25. My child does not agree with me and/or with other family members	0.140	0,064	0.757
28 I did not provide the best medical treatment for my child that I could afford	0.098	−0,125	0.718
17. The approach of the doctors hindered my explanation of the difficulties my child is experiencing	−0.109	0,110	0.653
7. We have been weakened as a family because of this experience	−0.100	0,106	0.646
12. In caring for the condition, I do not look after myself	−0.368	−0,088	0.416
15. The health status of my child is affecting my relationship with them	−0.013	−0,064	0.340
13. I can not imagine having more children because of my child's cleft	0.264	0,169	0.335
Cronbach's α coefficient	0.907	0,897	0,624

Table 6
Distribution of individual scores by parent's domains.

Group	N	Arithmetic mean	SD	Min	Max	Percentile		
						25.	Median	75.
Factor: social health of parents	73	40.63	23.47	0.00	88.64	23.86	38.64	59.09
Factor: social health of child	73	37.74	22.89	0.00	90.00	21.25	32.50	55.00

Table 7
Correlations of scores of individual quality life domains with socio-demographic parameters in the parent group.

		Factor: Social health of child	Factor: Social health of parents	Age	Gender
Factor: Social health of child	Correlation coefficient	1.000	0.474	0.060	−0.014
	P		< 0.001	0.470	0.886
Factor: Social health of parents	Correlation coefficient	0.474	1.000	0.047	0.048
	P	< 0.001		0.570	0.627
Age of parents	Correlation coefficient	0.060	0.047	1.000	−0.153
	P	0.470	0.570		0.121

in the current study. Parents are often unaware of how the behaviour other children may be detrimental to the development of their own child with a cleft; therefore, they do not attach great importance to it, or consider that the cleft is not the cause [32]. In fact, results of our previous study showed that adolescents with a treated cleft were much more burdened by the opinion of their peers than was assumed by their parents [14]. The views of parents of their own child's social health are very important because they feel that they know their child best, and their child will entrusted to them rather than those involved in their treatment. As parents must legally be present through the entire treatment procedure, their perception of the psychosocial health of their child is considered to have diagnostic value [16]. Hunt et al. [21] found that parents of children with clefts reported reduced self-confidence in the child, anxiety with appearance, shyness, difficulties in contacts with the opposite sex, and dissatisfaction with their appearance and speech. These results were consistent with the current study, considering the answers to items 23,24,27,26,18 and 20, which had average scores of under 3.

Despite the fact that the results of this current study confirmed that the quality of life of parents was not disturbed, that of children, as perceived by the parents, was strongly influenced by the opinions of the children. Social health is an essential and specific part of quality of life that is dependent on oral health. It is also affected by the importance of physical appearance imposed by society. Therefore, in adolescence, persistent mental disabilities can result if parents and therapeutic teams ignore these factors.

5. Conclusion

In the current study we created a specific instrument, QLPACA. This questionnaire comprised two subscales concerned with the social health of parents/respondents, and with the social health of adolescents as perceived by the parents. The questionnaire was valid and reliable, and it showed a satisfactory quality of life among parents of adolescents with clefts.

Disclosure of interest

The authors have not supplied their declaration of competing interest.

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