

# The relationship between congenital cleft lip and palate malformation, skeletal and dental occlusal anomalies, and the influence of its treatment on affected patients' oral health-related quality of life (OHRQoL)

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## SUMMARY

**Objective.** Non-syndromic (N/S) cleft lip and/or palate is the most common congenital anomaly to undergo long multidisciplinary treatment. This study investigates patients living with orofacial anomaly and having prolonged treatment by evaluating OHRQoL. It is essential to understand its influence on affected patients' daily life to prepare the most appropriate care for the complete physical, mental, and social well-being of an individual.

**Materials and methods.** In order to evaluate the influence of undergoing treatment and living with an orofacial anomaly on patients' OHRQoL, randomly selected patients with N/S cleft lip and (or) palate treated in the Clinic of Orthodontics of Lithuanian University of Health Sciences filled a modified CPQ questionnaire. The questionnaire of 60 closed questions was about undergoing treatment and its effect on patients' self-confidence level, mastication, fluent pronunciation, discomfort, ability to socialize, emotions, etc. Statistical data was expressed as a frequency and percentage.

**Results.** Of the 20 patients 13-24 year-old (mean age  $16.2 \pm 3.25$ ) 50% were males, 50% were females, 80% wore orthodontic braces, other 20% wore Quadhelix appliance. All 20 patients experienced pain during treatment, 60% of all patients avoided being among people, were bullied. 65% of respondents felt more confident and happy after orthodontic treatment began. 70% of all patients experienced difficulty in speaking, biting hard food.

**Conclusion.** The questionnaire showed no difference between different orthodontic appliance types and patients' OHRQoL, although overall prolonged treatment and orofacial anomaly does affect a patient's regular daily functions, oral hygiene habits, self-confidence level, social life, and mental well-being.

**Key words:** Cleft lip and palate, orthodontic treatment, OHRQoL, dental occlusion.

## INTRODUCTION

According to previously conducted studies, congenital anomalies are the most common cause of death among newborns (1). Almost eight million infants with congenital anomalies are born per year, which makes 6% of all infants' births in the world. (2). Cleft lip and/or palate are considered to be the most common congenital craniofacial malformation. 30% of oral clefts are associated with other major anomalies or specific syndromes, while 70% remain non-syndromic (3-5). Based on Linkevičienė L, congenital orofacial malfor-

mations among all congenital anomalies in Lithuania, rank from 6th to 7th in frequency (6). Most of the embryonic facial structures develop during the 4th and 8th weeks post-fertilization. Craniofacial anomaly, such as cleft lip and palate, may develop between the 7th and 12th week of gestation as a result of an impaired fusion of the embryonic prominences (7). In week 5 or 6 of a healthy pregnancy, maxillary prominences of the first pharyngeal arch fuse with frontonasal (medial) prominence and form the upper lip (8). In the case of structural fusion failure, the cleft lip occurs. According to the academic literature, clefts of the hard and soft palate are formed when the lateral palatine shelves fail to merge (9). Several studies have found a correlation between impaired migration of mesenchymal cells from the neural crest to the prominences

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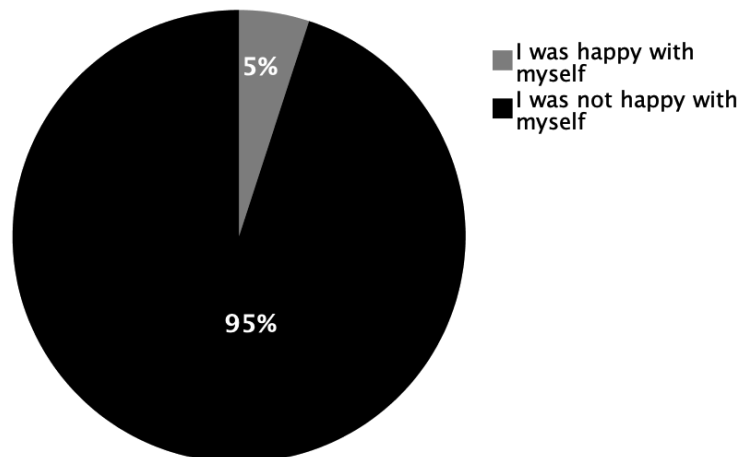
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and cleft lip and palate appearance. Other studies suggest that assisted reproduction, smoking, and decreased amount of folic acid consumption during pregnancy may be the main risk factors for developing cleft lip and palate malformation (7). It is important to acknowledge that an anatomical defect of the upper lip and palate affects not only the diet, facial appearance, speech, hearing, the psychological condition of a child, but also the development of the jaw and teeth (10, 11). It is common to observe tooth anomalies in children with non-syndromic cleft lip and/or palate, such as supernumerary teeth, hypodontia, microdontia, hypoplasia, rotations, transpositions in the cleft area (5). According to a study conducted by Rocha R. and co-authors, cleft lip and palate patients have specific characteristics, such as midface deficiency, Angle Class III malocclusion, and occasionally oronasal fistulas (12). Crossbite is commonly diagnosed due to the constriction of maxillary arch dimensions (13). Crossbite is associated with masticatory muscle thickness and activity reduction, as well as asymmetric mandibular movements (14). Cleft palate, surgical trauma, and scar tissue formation after surgery are common factors for Angle Class III malocclusion development (15).

One of the most common dental anomalies in the cleft lip and palate patients is the congenital absence of maxillary lateral incisors on the side of the anatomical defect (10, 16). Supernumerary teeth, particularly lateral or central incisors in the primary dentition, are evidenced in patients with cleft lip only (10, 17). Patients with cleft lip and palate are more prone to dental caries due to enamel hypoplasia in the central incisors. (10, 18). Microdontia and peg-shaped maxillary lateral incisors are other dental anomalies, common in the affected patients (18, 16).

The management of this malformation requires prolonged multidisciplinary treatment and highly specialized professionals. Therefore, early diagnosis of orofacial anomaly and the execution of precise intervention techniques are essential in order to prevent the development of malocclusion and dental defects. Due to the demanding treatment methodology, previously conducted studies have examined what challenges patients with orofacial anomalies endure in their daily lives. Several different tools, such as child perceptions questionnaire (CPQ) and oral health impact profile (OHIP), have been created by various researchers to evaluate oral health-related quality of life (OHRQoL) of patients with cleft lip and palate. The main focus of the questionnaire was to evaluate patients' oral symptoms, functional limitations, emotional and social well-being. (19) Having

**What was your opinion about yourself before the orthodontic treatment?**



**Fig. 1.** Patients' opinions about themselves before starting the orthodontic treatment

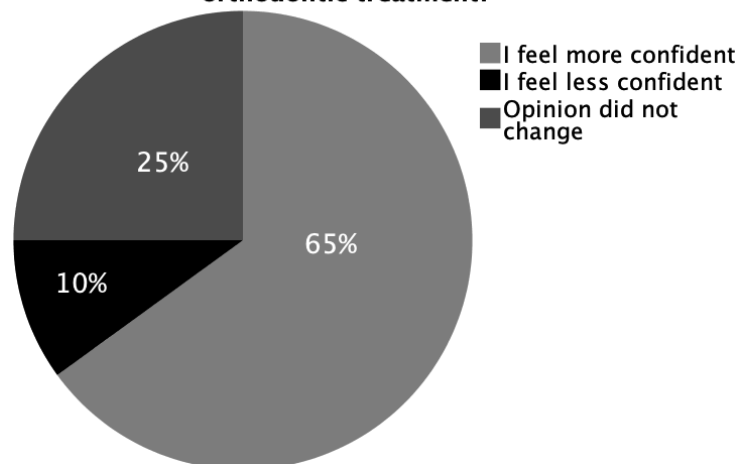
assessed what effect the treatment with different fixed orthodontic appliances has had on OHRQoL of cleft lip and palate patients, may help to ensure a better approach towards an individual's overall well-being. To achieve this goal, the study investigating patients living with orofacial anomaly and undergoing prolonged treatment by evaluating OHRQoL was performed.

## MATERIALS AND METHODS

Twenty randomly selected patients diagnosed with non-syndromic cleft lip and/or palate, treated in the Clinic of Orthodontics of Lithuanian University of Health Sciences, participated in the study. All the procedures used in the study were approved on the 7th of January, 2020, by the Lithuanian University of Health Sciences Ethics Committee (permit number BEC-OF-78) and allowed by the guardians of the participants'. A specific tool for evaluating the impact of different orthodontic treatment on affected patients' OHRQoL was created by modifying CPQ and OHIP questionnaires. The survey consisted of 60 closed questions, regarding undergoing treatment and its effect on patients' self-confidence level, mastication, fluent pronunciation, discomfort, ability to socialize, emotions, physical activity. The created tool was divided into separate parts of questions: 4 questions for the sociodemographic part, 12 questions for evaluating current overall health, seven questions for surveying the difference of dental hygiene habits, 23 questions for evaluating patient's feelings about ongoing treatment. The remaining 14 questions reflected the patient's emotional and social adaptation during orthodontic treatment.

All of the questionnaires were distributed to the participants in the Clinic of Orthodontics, Lithuanian

### Did your opinion about yourself change after starting orthodontic treatment?



**Fig. 2.** Patients' opinions about themselves after starting orthodontic treatment

University of Health Sciences. Statistical data was expressed as a frequency and percentage.

## RESULTS

The study included cleft lip and/or cleft palate patients aged 13-24 years (mean age  $16.2 \pm 3.25$ ). Of the 20 patients, 50% were males, 50% were females, 80% wore orthodontic braces, other 20% wore a Quadhelix appliance.

In the overall health before the orthodontic treatment assessment part, fourteen patients (70%) declared having experienced discomfort due to the congenital orofacial malformation. Such discomfort included gingival bleeding, bad breath, ulcers presented in the oral cavity. For the same 70% of patients, it was difficult to eat hard food such as apples and nuts, they ate food longer than other people, and had difficulty pronouncing particular words. In their oral hygiene habits assessment part, 12 patients (50% males, 50% females) answered that they brushed their teeth more than once a day, while the remaining 8 patients responded positively to brushing their teeth only once a day. Half of all the patients (7 males, 3 females) stated to never floss, yet 18 patients (90%) indicated to rinsing their mouth frequently.

Before the treatment, patients were not happy with themselves (Figure 1). All twenty patients experienced pain during orthodontic treatment. However, the majority of patients (65%), disregarding their malformation type, felt more confident and happy after the orthodontic treatment began (Figure 2). The pain that patients experienced was mostly mild, time of appearance varied. Dental hygiene habits improved for all 20 patients. The respondents indicated that they started to brush their teeth more often after the orthodontic treatment began. Out of 20 patients, 12 (6 females, 6

males) responded that they avoided reading or speaking in front of the class, sometimes felt sad and ashamed, constantly worried about their oral health. The same group of patients reported being bullied. All of the female respondents and only half of the male respondents often talk to their guardians about the social problems they experience, although all of the participants responded positively about having strong family support.

## DISCUSSION

Cleft lip and/or palate malformation is often diagnosed at an early age, causing young patients' frustration towards its management and how the future of their daily life may be affected. Although most of these particular orofacial anomalies are non-syndromic, multidisciplinary treatment is necessary in order to restore a correct skeletal pattern, prevent or manage dental defects, re-establish healthy, determined, anatomical structure development. As seen in the results of the study, cleft lip and/or palate patients do experience changes in their routine. The nasal, oral, and pharyngeal structures, which take part in speech producing, are the nose, nose cavity, lips, oral cavity, hard palate, velum, uvula, pharynx, Eustachian tube. These structures can be affected by cleft and do a great influence on inappropriate speech production, also limiting their musical or vocal capabilities. The normal anatomy of such structures is essential in order to correct speech inadequacies. In addition to the orofacial anomaly, having a combination of dental defects and skeletal problems like mandibular prognathism or maxillary hypoplasia leads patients to experience difficulty in verbal qualities, which constrains patients in their social life and causes negative performance of daily activities. This can negatively affect personality development.

Previously conducted studies showed that N/S cleft lip and/or patients indeed performed lower on measures of expressive language and verbal memory than healthy control groups. (6) There are many cleft speech defects, and in many literature sources, they are called differently. Even so, the hypernasality, various types of consonant articulation errors, and nasal air emission are the most common problems. The main predisposing factor of speech defects is velopharyngeal dysfunction. Little research has been done since the late 1980s on cognitive functioning in this group of patients. There is evidence that untreated posterior crossbite can cause facial asymmetry and may have an impact on the quality of life from a functional as well as an aesthetic point of view. (20) Having dental anomalies, such as hypodontia and microdontia, especially in the esthetic zone (any dentoalveolar segment that is

visible upon full smile from an objective standpoint) might decrease patients' wish to smile. Cleft lip and/or palate, in relation to supernumerary teeth and skeletal inadequacies, usually need surgical intervention, which might make patients more anxious.

A favorable outcome is patients' attitudes towards the treatment process. Even though they felt pain during orthodontic treatment, the patients' responses from the current study reflected that after the treatment began, they felt more confident and happy. Strong family support might have been an essential factor in this case. A positive alteration in patients' oral hygiene habits was indicated. Patients were motivated to develop better care of their oral health.

Studies with this particular group of patients are important in order to educate doctors of different specialties about developing better communication with the patient. As seen in the results of the study, cleft lip and/or palate patients are in need of understanding and support. A doc-

tor that manages the treatment should be well aware of what affected patients' experience and encourage them positively in every stage of the treatment. An active healthcare professional's interaction with the patient may change their point of view towards the anomaly's impact on OHRQoL. A team of different specialists should be in close communication in order to ensure the most qualitative treatment methods for the cleft lip and/or patients.

## CONCLUSION

The study showed that orofacial anomaly, dental and skeletal malocclusion do affect patients' social life and mental well-being, although no significant difference between different orthodontic appliance types and patients' OHRQoL were found. Positive outcomes are expected of studies that analyze cleft lip and (or) palate patients' OHRQoL; therefore, investigations should be continued.

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