

Condition of oral tissues in children with congenital cleft lip and palate

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ABSTRACT

Aim: To study the condition of oral tissues in children with congenital complete cleft lip, alveolar process, hard and soft palate.

Materials and Methods: From the examined 470 children National Specialized Children's Hospital "OKHMATDYT" (Kyiv, Ukraine) with congenital cleft lip and palate was analyzed: 302 patients aged 8-18 years were subject to in-depth analysis for clinical and radiological – 192 with unilateral and 110 with bilateral complete cleft lip, alveolar process, hard and soft palate.

Results: The average value of primary adentition in patients with unilateral and bilateral complete combined cleft is 69.53%, but in females this indicator is higher and in unilateral cleft 92.18% for female against 53.17% for male. In females with unilateral cleft retention – 40.62% and overcomplete – 10.93%. The same high indicators in bilateral cleft: retention – 36.58% and overcomplete – 12.19%. Retention and overcomplete have higher values for men – 44.93% and 23.19%, respectively. Chewing efficiency in females with bilateral cleft as a result of primary dentition is below 80%. Affected by caries – 90.73% in both groups. Inflammatory processes in the periodontal tissues are revealed (80,75%): chronic catarrhal gingivitis, chronic hypertrophic gingivitis, chronic generalized periodontitis. Manifestations of atopic and angular cheilitis in 39.09% and 23.63% with bilateral cleft lip and palate, glossitis in 29.09%.

Conclusions: Patients with congenital complete cleft lip, alveolar process, hard and soft palate have high rates of adentia, retention, overcomplete dentition and a wide range of diseases of the oral cavity, which negatively affects surgical and orthodontic rehabilitation.

KEY WORDS: caries, congenital defects, cleft lip and palate, defects of the dentition, diseases of periodontal tissues, diseases of the mucous membrane of the oral cavity

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INTRODUCTION

It is known that patients born with orofacial malformations, due to unfavorable anatomical and morphological changes, tend to be more susceptible to poor oral health compared to the general population [1, 2]. Previous studies have shown that among 4-6 year old children with congenital cleft lip and palate, the prevalence of dental caries reaches 71.9%. Anterior open bite, increased oral cavity damage and periodontal disease are also more common among children with cleft lip and palate [3, 4]. At the same time, different types of congenital cleft lip and palate, according to experts, are quite different in relation to dental diseases. For example, Hazza'a et al. concluded that patients with bilateral cleft lip and palate experienced more dental caries and poor oral hygiene than patients with unilateral cleft lip and palate [5].

There are a number of hypothesized factors that may explain increased risk of caries in children with cleft lip and palate compared with their unaffected peers. According to the authors, children with cleft lip and palate are usually born in low- and moderate-resource

socioeconomic areas [6, 7]. Parents typically have low oral health literacy regarding children with cleft lip and palate, also there is poor interaction with health care providers together with poor oral hygiene practices at home [8, 9]. In addition, infants with cleft lip and palate are bottle-fed longer on high-sugar formulas compared to breastfeeding [10, 11]. It was found that 55% of children with cleft lip and palate have decrease in salivation with a normal rate of salivary secretion (0.7 ml/min) [12]. Mouth breathing is common in patients with cleft lip and palate and can be considered as caries risk modifier due to a drying effect that reduces salivary production [8]. Finally, there may be a relationship between the anatomical features of the repaired cleft lip and palate, where scar tissue and uneven teeth can affect access to parts of the mouth and lead to prolonged retention of food debris [8].

Many researches have shown that oral microflora in patients with cleft lip and palate is associated with an increased frequency of potentially pathogenic fungal and bacterial colonization, particularly *Candida* species,

Table 1. Study of the adentia frequency in children with congenital complete cleft lip, alveolar process, hard and soft palate

Gender of the patient	Adentia of the central and lateral incisors								Primary adentia of other teeth	Secondary deformation	Full set of teeth	Total primary adentia	
	Partial adentia				In total	Multiple adentia							In total
	1 tooth	2 teeth	3 teeth			4 teeth	5 teeth	6 teeth					
Female	Unilateral cleft lip and palate (n=64)	57,81% (37)	10,93% (7)	4,68% (3)	73,44% (47)	6,25% (4)	-	1,56% (1)	7,81% (5)	10,93% (7)	-	7,81% (5)	92,19% (59)
	Bilateral cleft lip and palate (n=41)	17,07% (7)	24,39% (10)	17,07% (7)	58,53% (24)	9,75% (4)	2,44% (1)	-	12,29% (5)	4,88% (2)	9,76% (4)	14,43% (6)	75,60% (31)
Male	Unilateral cleft lip and palate (n=128)	33,59% (43)	7,81% (10)	0,78% (1)	42,19% (54)	-	-	-	-	10,16% (13)	1,56% (2)	46,09% (59)	53,17% (67)
	Bilateral cleft lip and palate (n=69)	21,74% (15)	24,63% (17)	4,34% (3)	50,72% (35)	5,80% (4)	2,90% (2)	8,69% (6)	17,27% (12)	8,69% (6)	2,89% (2)	20,28% (14)	76,81% (53)

Staphylococcus aureus, *Lactobacili* and *Streptococci mutans* and the formation of dysbiotic biofilm microorganisms [13, 14]. It has been shown that patients with cleft lip and palate have in 2.03 times higher risk of developing caries than children without cleft lip and palate [15]. The cariogenic and periodontogenic microbiota in children and adolescents with cleft lip and palate formed by microorganisms that considered to be highly pathogenic, inducing heavy course of the disease and complications in the postoperative recovery of patients. At the same time, biofilms on the surface of the teeth produce acids that demineralize the enamel and, ultimately, allow cariogenic bacteria to invade the enamel, dentin, and even the pulp [16]. The subgingival biofilm destroys the structural integrity of the epithelium, causing inflammation, which leads to the formation of periodontal pockets [17].

Regarding periodontal microbiota, *Campylobacter spp.*, *Fusobacterium spp.*, *Fusobacterium nucleatum*, *Prevotella intermedia/nigrescens*, *Parvimonas micra* and *Porphyromonas gingivalis* have been highlighted in patients with cleft lip and palate [18]. *Porphyromonas gingivalis* is not common in patients under the age of 18 and its presence in children and adolescents indicates immunological changes, due to microorganism locally affects periodontal tissue, avoiding the host's protective reactions. [19]. The above data suggest to implement an intensive preventive program that should be started at an early age, which may reduce the risk of periodontal disease in the future.

Thus, the current level of knowledge indicates that children with cleft lip and palate have more risk factors for poor oral health than the general population, and are also born with a variety of dental anomalies, which

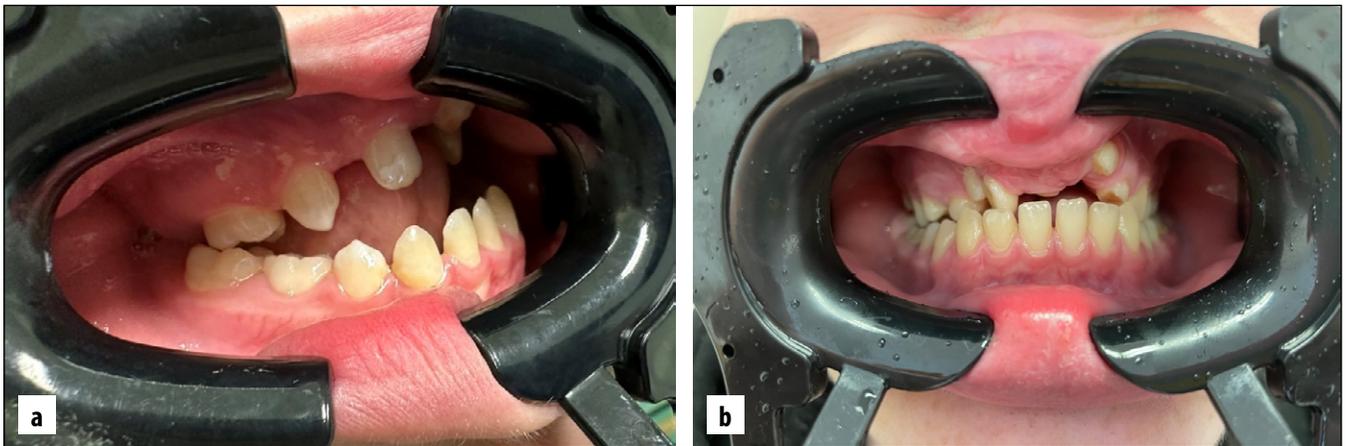


Fig. 1. a) Patient with congenital unilateral complete cleft lip, alveolar process, hard and soft palate. Multiple adentia; b) Patient with congenital bilateral complete cleft lip, alveolar process, hard and soft palate. Multiple adentia.

can not only increase susceptibility to caries, but also negatively affect periodontal condition.

Children affected by cleft lip and palate require multidisciplinary care to achieve optimal treatment results. Clinical command should prioritize oral health promotion activities to reduce the increased risk of dental disease, because early access to dental care can help to prevent the progression of dental, periodontal and oral mucosal diseases in the future.

AIM

To study the condition of oral tissues in children with congenital complete cleft lip, alveolar process, hard and soft palate.

MATERIALS AND METHODS

The dental status of 470 children with congenital cleft lip and palate was analyzed: 323 children with unilateral complete combined cleft lip, alveolar process, hard and soft palate, and 147 children with bilateral complete combined cleft lip, alveolar process, hard and soft palate. All children, according to their age, were operated on in the Department of Reconstructive and Plastic Microsurgery at National Specialized Children's Hospital "OKHMATDYT" (Kyiv, Ukraine) and undergo complex rehabilitation in the consultative and diagnostic polyclinic of National Specialized Children's Hospital "OKHMATDYT". 302 patients aged 8-18 years were subject to in-depth analysis for clinical and radiological examinations before the third stage of surgical rehabilitation (distribution by age corresponds to the stages of surgical rehabilitation): 192 with unilateral and 110 with bilateral complete cleft lip, alveolar process, hard and soft palate. Data were presented as absolute number and percentage. Data were analysed with the statistical package IBM SPSS Statistics Base (version 22).

RESULTS

From 470 patients, there were 323 (68.72%) children with congenital unilateral cleft lip and palate, 147 (31.27%) with bilateral. Gender – 109 (33.94%) female, 214 (66.23%) male. 302 (64%) patients were examined aged 8–18 years, and 192 (63.57%) from it had unilateral cleft lip and 110 (36.42%) had bilateral cleft lip. Left-sided cleft lip accounted for 202 (62.54%), of which 127 (59.35%) male and 75 (66.81%) female. Right-sided cleft lip accounted for 121 (37.46%), of which 87 (49%) male and 34 (31.19%) female.

From 323 unilateral – 60 (18.57%) from the capital of Ukraine and 263 (81.43%) from the regions of Ukraine, 203 of which patients (77.18%) with unilateral cleft lip and 60 (22.81%) with bilateral.

From 147 bilateral – 21 (14.28%) from the capital of Ukraine and 126 (85.71%) from the regions of Ukraine. From the capital – 8 female and 13 male, from the regions – 43 female and 83 male. Gender breakdown for bilateral cleft – 51 (34.69%) female, 96 (65.31%) male. From 110 patients, 23 (20.9%) came from the capital, 8 (19.51%) female and 15 (21.73%) male; 87 (79.09%) patients came from regions of Ukraine – 33 (80.48%) female and 54 (78.27%) male.

Maxillofacial deformities in the sagittal and transversal plane in congenital unilateral cleft lip and palate were found in 70.31% of children and the relative physiological norm was only 2.08%, and 73.26% in bilateral cleft lip and palate. Mainly, mesial occlusion with complications was observed in children with unilateral cleft in 34.37%, and in 34.65% with bilateral ones. Deep bite in 13.54% with unilateral cleft, and 10.89% with bilateral ones; open bite in 3.64% and in 5.94%. Crossbite observed in 35.94% and in 38.61% with bilateral cleft. A combination in two planes was found in 61.43% of all examined.

In children with congenital unilateral complete cleft lip, alveolar process, hard and soft palate only 7.8% of female patients have full set of teeth. In male patients, 1 tooth adentia – 33.59%, 2 teeth in 7.8%, 3 teeth in 0.78%, and

Table 2. Development pathologies and tooth eruption in children with congenital complete cleft lip, alveolar process, hard and soft palate

Indexes	Unilateral cleft lip and palate (n=192)		Bilateral cleft lip and palate (n=110)	
	Female (n=64)	Male (n=128)	Female (n=41)	Male (n=69)
Adentia	92,18% (59)	53,17 %	75,60% (31)	62,32% (43)
Retention	42,18% (27)	33,59%	36,58% (15)	44,93% (31)
Overcomplete teeth	10,93% (7)	21,13%	12,19% (5)	23,19% (16)
Combined pathology:				
Adentia + retention	26,76% (19)	6,61% (8)	26,83% (11)	11,59% (8)
Full set of teeth + retention + overcomplete teeth	-	3,30% (4)	14,63% (6)	-
Adentia + overcomplete teeth	2,81% (2)	1,65% (2)	-	2,9% (2)
Full set + retention	1,41% (1)	5,78% (7)	-	7,25% (5)
Full set + overcomplete	1,41% (1)	0,83% (1)	-	5,79% (4)
Adentia + retention + overcomplete	2,81% (2)	-	-	2,9% (2)

Table 3. The prevalence of the oral mucosa diseases in children with congenital complete cleft lip, alveolar process, hard and soft palate

Index	Unilateral cleft lip and palate (n=192)	Bilateral cleft lip and palate (n=110)
SD of DMFT	5,85	7,97
Angular cheilitis	22,91%	23,63% (26)
Glossit	26,56%	29,09% (32)
Atopic cheilitis	36,45%	39,09% (43)
Herpetic infection of the oral cavity	7,8%	13,63% (15)
Chronic recurrent aphthous stomatitis	19,79%	24,54%
Candidiasis	7,29%	10,0%

other teeth were primary adentia in 10.16%. In congenital bilateral complete cleft lip, alveolar process, hard and soft palate partial adentia stated 58.53%, and multiple adentia stated 12.29% in female. Only 14.43% of female patients have full set of teeth. In male patients partial adentia stated 50.72%, and multiple adentia stated 17.27% in male. Secondary deformation – 2.89%. 20.28% of male have a full set of teeth (Table 1, Fig.1).

In case of unilateral cleft lip and palate, the masticatory efficiency in two female cases was 78% – with adentia of the central lateral incisor and second premolars. 76% – adentia of the first and two premolars and a molar. The lowest level of masticatory efficiency in male – 80% of the adentia of the central incisor and second premolars in 2 cases. With bilateral cleft lip and palate, chewing efficiency as a result of primary adentia in 9 females is below 80%, and in combination with complete retention of teeth – in 4. Thus, 34.21% of the female have chewing efficiency below 80%. In 2 female, chewing efficiency is 64% and 68% as a result of secondary deformation. 2.9% male have chewing efficiency below 80%. Chewing efficiency of 76% was observed with adentia of 12, 22, 27, 35 teeth and 60% with dentition of 15, 17, 27, 37, 46, 47 teeth.

Patients with partial or multiple primary adentia need proper preventive and therapeutic support for the preser-

vation of teeth and periodontal tissues, taking into account the chosen method of replacing teeth with incomplete dentition. The number of anomalies in the position of the teeth on the cleft side is twice as large as the number of dystopias on the opposite side. The wide variability of retained teeth location in this pathology significantly affects the plan, tactics and terms of active and retention periods of orthodontic treatment (Table 2, Fig. 2). In patients with congenital bilateral cleft lip and palate, dystopia of incisors, canines, premolars and molars is more pronounced than in patients with unilateral defects.

One of the important stages of rehabilitation for children with congenital cleft lip and palate is monitoring the condition of the hard tissues of the teeth (Fig. 3). The indicators of the prevalence and intensity of the carious process among the patients examined by us were determined as quite high. The caries situation in children with congenital bilateral cleft lip, alveolar process, hard and soft palate is significantly worse. In patients with unilateral cleft, 13.54% (26 children) had intact teeth. 551 teeth in 166 children affected by caries on the upper jaw with unilateral cleft – 3.31 teeth in one patient; 422 teeth on the lower jaw. In total, the incidence of caries is 5.85. With bilateral cleft, 502 teeth on the upper jaw and 359 teeth on the lower jaw were affected by caries. In total, the incidence of caries is 7.97. 98.18% of children



Fig. 2. Patient with congenital unilateral complete cleft lip, alveolar process, hard and soft palate.

with bilateral cleft were affected by caries – only 1.82% (2 children) had intact teeth. Complicated caries in children with bilateral cleft palate was higher than with unilateral and stated 27%.

Non-carious lesions in the form of hypoplasia were observed (24.23%), mainly systemic hypoplasia that located on teeth of the same period of formation. Enamel hypoplasia was often accompanied by a change in color, tooth sensitivity, reduced acid resistance, and erosion.

Patients with cleft lip and palate need biofilm control due to difficult access to areas for conventional hygiene techniques, reduced quality of self-cleaning, mouth breathing, tendency to retain plaque, orthodontic treatment of complex bite deformities that can last 5-7 years or more. Even in patients with a strong motivation for hygiene, long-term fixed design elements lead to high bacterial insemiation, cause changes in the color of tooth enamel around braces, the formation of enamel defects, and hyperesthesia. Patients with both unilateral and bilateral cleft lip and palate complained of tooth hyperesthesia, which can also be caused by an inflammatory reaction in the pulp in reversible and irreversible pulpitis, periodontal diseases. Difficult hygiene of the oral cavity with non-unions at the same time complicates the fixation of bracket systems and orthodontic treatment in general. Even with the lowest values of the

digital data of the hygienic indices in these patients, the hygienic condition of the oral cavity was determined as unsatisfactory (72.35%), despite the constant control of the pediatric dentist, training in special care methods, and encouragement to use modern care devices.

The multifactorial nature of abnormalities of the maxillofacial system in congenital cleft lip and palate is a significant risk factor for periodontal disease. Bite anomalies increase the severity of morpho-functional disorders in the periodontium. Orthodontic treatment of patients with periodontal diseases leads to elimination of functional loads and restoration of chewing efficiency. Expanding the maxillary arch helps to change its arch shape and reduce crowding of teeth.

The condition of the periodontium in case of bilateral cleft lip and palate was much worse than in unilateral. These patients are more prone to deep periodontal destruction of the teeth adjacent to the gap. Healthy periodontium stated 20.31% with unilateral cleft and 18.18% with bilateral. These are patients with regular dental examinations and proper oral hygiene. Children with both unilateral and bilateral cleft had average severity gingivitis 55,75% та 57,27 %. Inflammatory processes in the periodontal tissues are revealed (80,75%): chronic catarrhal gingivitis, chronic hypertrophic gingivitis and chronic generalized periodontitis.

In these children with both unilateral and bilateral cleft was revealed predisposition to inflammatory diseases of the mucous membrane of the oral cavity. Manifestations of atopic and angular cheilitis in 39.09% (43 people) and 23.63% (26 people) with bilateral cleft lip and palate, and glossitis in 29.09% (32 people) (Table 3, Fig.3). A short frenulum of the upper lip, that contributes to the development of periodontal diseases, was found in children with congenital cleft lip and palate about a third of cases (31.12%), and much lower percentage of tongue frenulum abnormality (23.17%) and pathology of the upper jaw (27.81%).

DISCUSSION

The examined contingent of patients that operated at National Specialized Children's Hospital "OKHMATDYT" with congenital complete combined cleft of the maxil-



Fig. 3. Patients with congenital bilateral complete cleft lip, alveolar process, hard and soft palate (a,b,c).

lofacial region that have extremely complex anatomical disorders and interdependent functional disorders – chewing, swallowing, breathing, speech, hearing and smell. Surgical rehabilitation restores unfused anatomical structures that must be developed to fulfill the primary functional state and facial and smile aesthetics. The interstage postoperative period requires close cooperation with otolaryngologists, speech therapists, psychologists, dentists, orthodontists and specialists to strengthen the health of children with cleft who have pronounced comorbid pathology.

Passinato Gheller et al. found that gingivitis was observed in 52% and 29%, and mild periodontitis in 48% and 22% in groups with and without cleft lip and palate, respectively [2]. Similarly, Veiga et al. found that in individuals with and without cleft lip and palate, 49% and 75% of patients had good gingival health, 22% and 24% had localized gingivitis, and 29% and 1% had generalized gingivitis. The obtained results demonstrate that the presence of cleft lip and palate is a determining factor for a higher risk of gingivitis [20].

Plakwicz et al. [21] similarly to Wyrębek et al. analyzed data of 15 patients with bilateral cleft lip and palate and found heavy bleeding and loss of clinical level of gum attachment teeth adjacent to the cleft. [22].

Gaggl et al. reported that patients with cleft lip and palate are prone to deep periodontal destruction of teeth adjacent to the cleft lip and palate. The largest number of teeth in patients with cleft lip and palate showed mobility of the I stage. According to the authors' conclusions, a critical periodontal situation was found in patients with unilateral and bilateral cleft lip and palate [23].

Assessing gingival recession as a marker of periodontal health in patients with congenital cleft lip and palate (n=200), Almeida et al. reported that the presence of recession was high, as 75% of subjects in the sample had at least one tooth with gingival recession; and 100% of persons >43 years old had at least one episode of recession, and most recessions were I class according to Miller [24]. Stec et al. reported that gingival recession was found in teeth adjacent to the cleft area [25]. Contrary to these conclusions, Wyrębek et al. did not observe gingival recession in their patients with cleft lip and palate [22].

Patients with cleft lip and palate are born with a variety of dento-maxillofacial anomalies that may not only increase susceptibility to caries, but also negatively affect the periodontal condition. Al Jamal et al. stated that the absence of teeth was found in 66.7% of patients with cleft lip and palate; the most frequently missing tooth was the lateral incisor of the upper jaw. Supernumerary teeth were found in 16.7% of patients; 37% had microdentia; 70.5% taurodontism; 30.8% – trans-

position and/or ectopic teeth; 19.2% had dilacerations; 30.8% had hypoplastic teeth. [26]. Dens invaginatus is always found in the lateral incisor of the maxilla, creating a pathological dental pocket. Such pocket destroys the periodontal tissues and allows bacteria to multiply inside, which cannot be easily removed in patients with cleft lip and palate [27].

Deformation of the upper jaw, a large number of supernumerary teeth, and incorrect placement of teeth in patients with cleft lip and palate lead to crowding of teeth and malocclusion [28].

Considering that, age is positively associated with the progression of periodontal disease, and individuals with cleft lip and palate are more likely to have plaque accumulation and gingival inflammation, clinicians should increase preventive dental care for patients from an early age [29]. In addition, in infants and children with orofacial clefts, pre- and post-operative oral inflammation pose a main risk for wound healing disorders and failure of surgery [30].

The vast majority of patients with congenital cleft lip and palate complete multi-link, timely and consistent surgical and orthodontic treatment with total positive results by adulthood.

Rehabilitation of congenital defects of the maxillofacial area consists in the anatomical and functional restoration of disturbed structures and functions performed by the orofacial apparatus: breathing, chewing, swallowing, speech, hearing and smell. Children with congenital unilateral and bilateral complete cleft lip, alveolar bud, hard and soft palate need rehabilitation to a great extent.

Restoration of impaired functions occurs before adulthood, and continues in the adult period of life. In the rehabilitation interdisciplinary doctors team, main leading role belongs to orthodont, together with the maxillofacial surgeon and the pediatric dentist to implement fully coordinated, multi-stage and multi-component treatment, the basic principles of which are professionalism, timeliness and complexity based on the continuity of polyclinic dynamic supervision with a specialized inpatient department.

The results of the examination can be used to optimize treatment and prevention actions in a complex interdisciplinary approach to the rehabilitation of children with congenital defects of the maxillofacial area; to optimize the plan of orthodontic treatment at the stages of surgical treatment, creating sufficient space on the alveolar of missing teeth by moving or replacing missing teeth: mediat-prosthesis; adhesive bridge; installation of an implant or permanent prosthetics in the future. Final corrective treatment continues into adulthood, especially in female patients.

CONCLUSIONS

1. The average value of primary adention in patients with unilateral and bilateral complete combined cleft is 69.53%, but in females this indicator is higher – 16.18% (85.71% vs. 60.91%) and in unilateral cleft 92.18% for female against 53.17% for male. This indicates a functional, social and aesthetic problem and require professional, planned, consistent rehabilitation from doctors – an operating maxillofacial surgeon as a coordinator, a dentist, an orthodontist, and an orthopedist.
2. Orthodontic treatment of sagittal and transverse deformities is significantly complicated by defects in the development of teeth – adentia, retention – 40.62% and overcomplete – 10.93% in females with unilateral cleft. The same high indicators in bilateral cleft: adentia – 75.61%, retention – 36.58% and overcomplete – 12.19%. Retention and overcomplete have higher values for men – 44.93% and 23.19%, respectively.
3. Chewing efficiency in females with bilateral cleft as a result of primary dentition is below 80%, and in combination with complete retention of teeth is significantly reduced. Secondary deformation stated in 2.65% patients which indicates the insufficiency of timely preventive and rehabilitation work of dentists.
4. Affected by caries – 90.73%: with congenital unilateral cleft lip and palate – 86.46%, with decay-missing-filled teeth –5.85; with congenital bilateral cleft lip and palate – 98.18%, with decay-missing-filled teeth –7.97. The hygienic condition of the oral cavity was determined as unsatisfactory in the majority of patients of both groups. Healthy periodontium stated 20.31% with unilateral cleft and 18.18% with bilateral. In children with both one- and two-sided cleft, with a predominance of the average severity of gingivitis – 55.75% and 57.27%. Manifestations of mucous membrane diseases of the oral cavity were observed mainly cheilitis, glossitis, chronic recurrent aphthous stomatitis, herpes and candidiasis.

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CONFLICT OF INTEREST

The Author declare no conflict of interest

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