Modern aspects of interdisciplinary treatment in children with bilateral cleft lip and palate

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According to the World Health Organization incidence of cleft lip and palate ranges from 0.6-1.6 cases per 1000 newborns per year. Thus, bilateral full cleft lip and palate occurs less frequently around 15–25%. Purpose: to analyze treatment methods for children with bilateral cleft lip and palate during the period of a removable bite. A review of 51 literature sources from 1951 to 2019 was carried out. Occlusion development features in bilateral cleft lip and palate patients during mixed dentition period were analysed. Main anatomical features of the maxillofacial region in children with bilateral cleft lip and palate during mixed dentition period is narrowing of the upper and lower dental arches as well as presence soft tissues scars of the upper lip. The main methods of treatment for such children is surgical reconstructive operations, including the elimination of the anatomical defect of the hard palate using mucoperiosteal flaps from the lateral parts of the hard palate combined with orthodontic treatment methods. Thus, an integral method of treating children with bilateral cleft lip and palate during mixed dentition period is a comprehensive approach, including surgical reconstructive operations, as well as orthodontic treatment, the main purpose of which is to expand and extend dental arch, which results in normal occlusion therefore eliminating myofunctional disorders.

Key words: cleft lip, cleft palate, mixed dentition, occlusion features, orthodontic treatment

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Современные аспекты междисциплинарного лечения детей с расщелиной губы и нёба

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По данным Всемирной организации здравоохранения возникновение расщелины губы и неба составляет от 0,6 до 1,6 случая на 1000 новорожденных в год. Так, двусторонняя полная расщелина губы и неба встречается реже и составляет 15–25%. Цель исследования: проанализировать методы лечения детей с двусторонней расщелиной губы и неба в период сменного прикуса. Проведен обзор 51 литературного источника с 1951 по 2019 год. Проанализированы особенности развития окклюзии у пациентов с двусторонней расщелиной губы и неба в сменном прикусе. Основными анатомическими особенностями челюстно-лицевой области у детей с двусторонней расщелиной губы и неба в период сменного прикуса являются сужение верхней и нижней зубных дуг, а также наличие рубцов мягких тканей верхней губы. Основными методами лечения таких детей являются хирургические реконструктивные операции, включающие устранение анатомического дефекта твердого неба с помощью слизисто-надкостничных лоскутов из боковых отделов твердого неба в сочетании с ортодонтическими методами лечения. Таким образом, главным методом лечения детей с двусторонней расщелиной губы и неба в сменном прикусе является комплексный подход, включающий в себя хирургические реконструктивные операции, а также ортодонтическое лечение, основной целью которого является расширение и удлинение зубных рядов, что приводит к нормализации прикуса, а также к устранению миофункциональных нарушений.

Ключевые слова: расщелина губы, расщелина неба, сменный прикус, особенности прикуса, ортодонтическое лечение

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Bilateral cleft lip and palate (BCLP) is considered the most severe pathology among congenital malformations of the maxillofacial region. Depending on the severity of the pathology, the alveolar bone, hard, soft palate may be involved. Immediately after birth children with BCLP are liable for most serious complications, sometimes resulting in newborn mortality.

The presence of a bilateral anatomical defect of the lip, alveolar process of the upper jaw leads to the advancement of the intermaxillary bone forward and presents deformed position of the alveolar processes. This type of anomaly that occurs during the prenatal period. As a result of teratogenic factors affecting the fetus, anatomical and physiological disorders of the maxillofacial region occur.

Our treatment protocol in children with BCLP in the neonatal period includes pre-surgical orthodontic treatment using removable and non-removable orthodontic devices.

It takes two weeks, after which the primary cheiloplasty is performed.

Furthermore, a patient with BCLP is undergoing multidisciplinary treatment including various specialists (orthodontist, ENT, speech therapist, psychologist, social worker).

These treatment methods help normalize the vital functions of the newborn, such as breathing, sucking, and swallowing.

However, as a result of active growth patterns in a child up to the age of 6 years old, pathological occlusion develops in a child with BCLP

Children with BCLP require mandatory orthodontic assessment and treatment. BCLP affects not only aesthetic, but also functional features of the maxillofacial area.

When planning BCLP patient's treatment strategy, comprehensive diagnostics is vital. Its main purpose is to assess the profile of the child, state of occlusion, predict possible treatment outcomes and its effectiveness.

At present various treatment methods are being used. Surgical methods aim to eliminate secondary deformities, while orthodontic treatment is carried out using various fixed or removable, intra- or extraoral orthodontic appliances.

However, the effectiveness of these treatment methods was not compared with one another. As a result, no consensus exists on a unified multidisciplinary approach for the treatment of BCLP patients.

All things considered, comprehensive, detailed and phased approach to the methods of treating children with BCLP during a mixed dentition is still relevant today.

Analysis of 51 literature sources from 1951 to 2019 was carried out. The focus of analysis remained on assessing mixed dentition occlusion in children with BCLP. Following parameters such as teeth shape and dental arches, as well as sagittal and transversal plane changes in dentition, soft tissues of maxillofacial region and tongue position.

Treatment methods analysis in BCLP patients, such as surgical (reconstructive) operations aimed to eliminate secondary deformities; treatment with orthodontic appliances (removable plate devices, non-removable devices (Quad Helix, Hyrex, Mini-Hyrex, Haas, Fan-type Leone), non-removable equipment – braces (2 * 4); functional devices (facial masks, trainers, elastospositioners, Frenkel device), aimed to help myofascial area development, as well as fix and stabilize result achieved during orthodontic treatment.

Bilateral cleft lip and palate causes significant changes in osteoarticular system of the maxillofacial region. Consequently, during the

treatment process characteristic anatomical and functional disorders, which progress with the formation of a removable bite.

According to N.J.Prescott, S.Malcolm (2002), children with BCLP present narrowed transverse and longitudinal dentition, mainly in the area of the first permanent molars and canines on the upper jaw [1, 2]. N.A.Panahov et al. (2015) in their study observed narrowing in this particular area which doesn't lead to shift in the midline, however, narrowing of the dentition in the area of the first and second premolars of the upper jaw leads to shortening of the tooth segment length by 3.8 mm [3]. In her study O.I.Arsenina. (2017) emphasizes that the distance between canines of the upper jaw in children with BCLP decreases by 2.1 ± 0.4 mm in relation to the norm. As well as 1.8 ± 0.5 mm between the first upper molars [4].

Mesial displacement of dentoalveolar arch in the lateral segments, dystopia and palatine incline of the first and second premolar were observed in this study [4, 5].

E.Ya.Hasanli et al. (2015) assessed the degree of narrowing of the dentition. Their study showed that the ratio of dentition in central occlusion has been changed. Biometric analysis of the jaw models showed that with this pathology in mixed dentition in the position of central occlusion, the reverse incisal overlap is formed, as well as a bilateral cross bite [3].

The teeth closure is Engle class III in the area of lower canines and Engle class II in the area of lower permanent molars. This inclination and rotation in different directions is reflected in changes of the Spee curve [3, 6, 7].

Clinical studies carried out by A.G.Nadtochy et al. (2012) indicate that violations of the fissure-tubercle contact of the teeth lead to dysfunction of the lower jaw. Which doesn't allow for normal lower jaw movement in the sagittal, vertical and transverse planes [8].

Children with BCLP are subjected to various changes in the osteoarticular system. In addition to those, there are significant changes to the soft tissue of the maxillofacial area, such as an irregular shape of the upper lip, low and short attachment of upper and lower lip frenum, shallow vestibule of the mouth, as well as a compromised position of the tongue [8–11].

N.V.Starikova in her study (2014) assessed results of an ultrasound investigation. Which revealed an asymmetry of the muscle structures of the tongue, a violation of the configuration of the tongue with a low location of the apex and a high position of the tongue [12].

At the same time, multispiral computed tomography, showed a decrease in the size of the tongue and an increase in the amount of free space in the upper part of the oral cavity were detected. Consequently, these changes lead to secondary deformations (lower macrognathia, lingual inclination of the lower incisors, as well as retrogenia) [11–13].

According to A.V.Zernov (1997) children with BCLP in the period of mixed dentition have a considerable over-jet due to intermaxillary bone protrusion forward and down [14]. The incisors inside the intermaxillary bone are subjected to anomalies in shape, size and position. In the aforementioned study, 63% of children have supernumerary lateral incisors, 31% have a compromised shape of the lateral incisors ("spike-shaped"), and 6%, have lateral incisors which are located within the boundaries of the anatomical cleft.

If the eruption and the position of the permanent anterior and posterior teeth are disturbed, the occlusal plane is deformed: a bilateral cross bite, narrowed V-shaped upper dentition follows [14]. In her study, A.O.Oleinik (2014) noted the asymmetric dentition in the max-

illa, which progresses with growth of the maxilo-facial area [15] combined with slow growth of the maxilla in the sagittal direction [15–17].

Clinical and scientific studies of the maxillofacial area development features in children with BCLP in mixed dentition period emphasize the severity of deformations in both maxilla and mandible. Therefore, treatment methods require structured interdisciplinary approach.

At present, a lot of methods for the treatment BCLP during mixed dentition period exist. Surgical protocol includes reconstructive operations, the purpose of which is to restore the continuity of the alveolar arch, stabilize fragments of the upper jaw, accelerate the eruption of permanent teeth located in the defect area, and eliminate the oronasal communication.

Professor Ad.A.Mammedov (2012) proposed the elimination of a defect of the anterior hard palate using a flap from lateral part of the hard palate. The flap then is distributed along the plane of two mucoperiosteal flaps [17].

This method eliminate defect in the middle part of hard palate by detachment of tissues, moving along the plane and overturning the mucoperiosteal and muscular-muscular flaps, suturing of the muscular groups of the palatopharyngeal ring in the area of the defect as well as narrowing palatopharyngeal ring [18, 9]. These methods reduce time of medical, psychological, pedagogical and social rehabilitation of children and accelerate the recovery of impaired speech and socialization of a child with BCLP [20, 21].

H.Schweckendiek (1951) proposed reconstructive surgery of hard and soft palate in two stages: first stage is staphyloplasty (soft palate) at 7–8 years, and the second stage is uranoplasty (plastic within the hard palate) at 12–15 years old [22]. According to the author, the early restoration of the soft palate contributes to the proper development of speech, and at the same time does not adversely affect the development of the middle zone of the face, and there is no violation of the growth and development of the upper jaw.

However, in our opinion, such late primary veloplasty does not contribute to better speech development. In our practice, we believe that an earlier restoration of palatopharyngeal ring facilitates speech development.

S.N.Bessonov (2007) conducted early secondary rhinocheilognatoplasty. This method enables normal development of the middle part of the face and reduces the severity of secondary deformities [23, 24]. A mucous membrane flap in being cut out on both sides, forming the lining of the nasal base, dissection of circular muscle of the mouth, mobilizing lip fragment and wing of the nose at its base from the upper jaw, filling the bone defect with a lyophilized bone graft. However, in some children, the volume of bone tissue in the area of transplantation is not sufficient for the eruption of lateral incisors [24].

Another, more successful technique is uranoplasty suggested by G.Kotov. and I.V.Muratova (1999), optimized form by P.Tokarev (2019). Excision mucous membrane strip is carried out in the tongue area, which allows to adapt the edges of the wound to each other more accurately and reduce the formation of scar tissue. [25, 26]. The main objective of this technique is a short postoperative period, which makes it possible to reduce the length of hospitalisation and speed up the restoration of speech through speech therapy and physiotherapy. All this aims to stimulate muscles of the face and oral cavity.

Rehabilitation of children with BCLP after reconstructive surgery is a complex process. As well as surgical reconstruction, children

with BCLP undergo orthodontic treatment. With this pathology, a significant narrowing of the dental arch is present. Orthodontic treatment is aimed to primarily expand maxillary dental arch. Various removable and fixed orthodontic appliances are used.

Removable orthodontic appliances are aimed to expand maxillary dental arch by transversally displacing the canines of the upper jaw, as well as the first permanent molars.

N.A.Vorozheykina (2019) proposes the use of single-jaw removable plate appliance [27]. This appliance is a plastic base plate with clasps, with(?) longitudinal sectoral cut and a mechanical orthodontic screw between the sectors. The movement of canines and the first permanent molars of the maxilla is carried out due to the activation of the orthodontic screw. In addition, the movement of the central and lateral incisors of the maxilla occurs vestibularly by activating the traction spring. The advantages of this device is that it is small and does not have massive occlusal pads.

In order to correct the cross occlusion in the lateral and anterior sections of the dentition, mandibular functional guiding devices are used. These devices can also eliminate blocking occlusal contacts, which makes it possible to move the mandible to a central position [27].

In case of reverse incisal overlap (up to 1.5 mm), it is necessary to lengthen the upper dentition using removable lamellar devices fixed on the teeth using clasps. Lengthening of the dentition in the anterior region occurs when orthodontic screw is activated. In turn, the mandibular functional guiding devices aim to reducing depth of the incisal overlap. Frontal and lateral sections of the occlusal plates allow to unlock maxillary incisors. This creates freedom of movement in the mandible in the anterior and posterior directions in the sagittal plane [28].

Quad Helix devices are highly efficient when growth of the maxillary dentition in the transversal and sagittal directions has to be optimized. This unit is a four spiral wire with a base with a metal screw and ring hooks. The whole system exerts continuous mechanical pressure on the maxillary dentition and expands it. This system significantly normalize position of the maxillary bone by correcting position of the upper incisors and simultaneously symmetrically expands maxillary dentition [29, 30].

In their research, M.R.Vasant and S.Menon (2009) emphasize that Quad Helix is the optimal alternative to Rapid palatal expansion (RPE) devices because it can correct the rotation of molars [31]. pressure before any movement of the teeth or physiological correction of the suture [32].

Effective devices for rapid expansion of the palate (RPE) are Hirex (Hyrex), Haas (Haas) and Fan-type Leone [33].

In his work, J.A.Freitas (2012) notes that expansion using Hyrex is carried out with hyper correction so that the palatine cusps of the upper molars and the buccal cusps of the lower molars are at the same level [30]. Hyrex occurs on permanent molars and premolars of the upper jaw; support wire elements can also be added to correct the stiffness of the apparatus.

Hyrex has several advantages such: it does not irritate the mucosa of the palate, allows to maintain oral hygiene at a satisfactory level, and also makes it possible to use the device for extended periods of time in case of late eruption of premolar by activating screws in intraoral access. HoOn the other hand, it does not allow to achieve the maximum vestibular deviation of the fixing teeth, in comparison with the Haas. It also does not have an acrylic stand on the palate

due to this trauma to the mucosa of the tongue, as well as profuse salivation and constant discomfort.

Also, Hyrex should be in the oral cavity after optimal expansion is achieved for another 3–5 months to re-establish the middle palatine suture [30, 33–35].

Daniel Santos Fonseca Figueiredo and Lucas Cardinal (2016) developed Mini-Hyrex, which is designed with a mini-screw located in the front area of the dentition. This reduces the negative impact of orthodontic treatment in children with BCLP [36]. Studies show that Mini-Hyrex minimizes difficulties in maintaining oral hygiene, and is also aimed at more effective expansion in the anterior region, while limiting expansion in the posterior region [37].

A similar device for rapid palatal expansion is Haas. The distinctive feature and the main advantage of its design are fixed orthodontic rings and mounts on the temporary second upper molars and temporary cuspids, while there is no mechanical aggressive effect on the permanent teeth.

In the case of a significant narrowing of the maxillary dentition in the form of a V-shape, it is rational to use the Fan-type Leone device. Umarevathi Gopalakrishnan (2017) recommends use of the Fan-type Leone apparatus with the goal of expanding only front of the palate, deactivating the device immediately after reaching the necessary expansion of the palate, which prevents hyper-healing [38].

Professor O.I.Arsenina (2015) proved the clinical effectiveness of early orthodontic treatment in children with dentoalveolar anomalies in the period of temporary dentition on a fixed 2 × 4 braces system [39]. Mechanism of these structures aims to correct the pathological shape of the dentition due to weak and constant forces that lead to oblique-rotational and body movements of the teeth. The use of these fixed orthodontic appliances is shown in 7–9 year olds with distal occlusion, reverse incisive disocclusion, and wrong position of individual teeth. The 2 × 4 technique consists of 2 orthodontic locks (tubes) fixed on the first permanent molars and 4 braces on the central and lateral maxillary incisors.

It should be noted that for orthodontic correction in children with distal occlusion in combination with fixed orthodontic systems, extraoral orthodontic appliances in the form of facial arches should be used. Such a technique for treating dentoalveolar anomalies eliminates dentoalveolar pathology as well as sagittal fissure and improves facial features [4, 39].

To normalize the myofunctional facial complex, it is recommended to use facial masks (Petita, Delara, Tubinger). These masks consist of a metal frame and two supports fixed to the child's forehead and chin. Masks should be used in cases of mesial occlusion, when the mesial displacement of teeth 3.6, 4.6 is 0.5–1 tubercle and the back incisal overlap is combined with a sagittal fissure of up to 2 mm [40]. So, S.Dogan (2012) in his study demonstrated the effectiveness of facial masks for children with cleft lip and palate [40, 41].

It should be noted that children with congenital pathology of the maxillofacial area require a systematic, consistent, comprehensive approach, since with this pathology significant myofunctional disorders are present, which leads to impaired functions such as swallowing, chewing, pronunciation and malocclusion [42].

In order to correct myofunctional disorders, it is advisable to use myotrainers. E.A.Shatova (2019) argues that the use of the INFANT trainer is necessary from the time of eruption of temporary dentition (up to 7 years). This trainer helps to form correct shape of the palate and nasal passages, which improves the quality of sound pronunciation

and intonation [43]. From the age of eight it is recommended to use the T4K trainer, which combines the properties of a functional simulator and positioner. Thus, muscle function is normalized due to the removal of excessive muscle tension and pressure on the dentoalveolar part [44, 45].

An alternative to preorthodontic trainers is elastopositioners. I.V.Tokarevich (2012) proposes the use of "LM-Activator" for children in temporary dentition. This unit is soft enough to be comfortable to wear, and at the same time tough, so that the child cannot damage it [40]. Numerous clinical and scientific studies by O.I.Arsenina (2013) demonstrate the effectiveness of the OE Corrector elastopositioners for treatment of deep disocclusion, vertical disocclusion, distal occlusion, retrusion and protrusion of incisors, lower jaw displacement, as well as for training the muscular complex of the maxillofacial region and stabilization of the treatment result [46].

To normalize the position of the tongue in the oral cavity and prevent the lower jaw from moving forward in children with BCLP during the period of a temporary dentition, it is recommended to use a type III Frenkel device [47–50].

Friedy Luther and Zarana Nelson-Moon (2013) found in the book Orthodontic Retainers and Removable Appliances: Principles of Design and Use that each component of this functional retainer plays a key role [49].

As an example, the buccal plate is designed to reduce the load of the buccal muscle on the upper dentition, helps to increase the upper jaw width; acrylic labial pads affecting the circular muscle of the mouth displace the upper incisors into protrusion, while the labial arch fixed on the incisors of the lower jaw promotes the displacement of these teeth into retrusion [51].

Based on current literature review it is clear that main anatomically children with bilateral cleft lip and palate during the period of temporary dentition have severe narrowing of the maxillary dental arch, also complicated by soft tissue scarring after surgery.

In view of this, it has been proved that surgical reconstructive operations, as well as complex interdisciplinary orthodontic treatment using removable and non-removable devices, the main purpose of which is to create the correct aesthetic and functional occlusion, are integral methods of treating such children.

In addition, the effectiveness of the use of functional orthodontic appliances has been demonstrated, the efforts of which are aimed at eliminating myofunctional disorders.

It has been established that the use of a combination of extraoral and intraoral orthodontic appliances allows to eliminate dentoalveolar pathology, improve facial features, and eliminate the sagittal gap.

Clinical and scientific studies demonstrate that the implementation of complex surgical and orthodontic treatment can reduce the rehabilitation time for children with BCLP, thereby achieving a stable aesthetic and functional result.

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