GENERAL REVIEW

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Orthodontic therapy in children with bilateral cleft lip and palate: presurgical orthopedic aspects in the newborn period

Ortodontska terapija kod dece sa bilateralnim rascepom usne i nepca: prehirurški ortopedski aspekti u periodu novorođenčeta

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Ključne reči:
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Introduction

Bilateral cleft lip and palate (BCLP) represent the most severe clinical form of clefts. A child born with BCLP has a characteristic facial appearance that is noticeable immediately upon birth (Figure 1). The clinical picture is variable, and the degree of deformity can vary from mild to severe; the protrusion of the premaxilla determines the degree of its severity. Regardless of the morphological variations within this type of cleft, the clinical picture is always dramatic and severe [1] (Figure 2). BCLP are reported with a 9.2% incidence of all clefts [2]. Within this group of clefts, there is a certain

Fig. 1 – Facial appearance of one-day-old newborn with bilateral cleft lip and palate.

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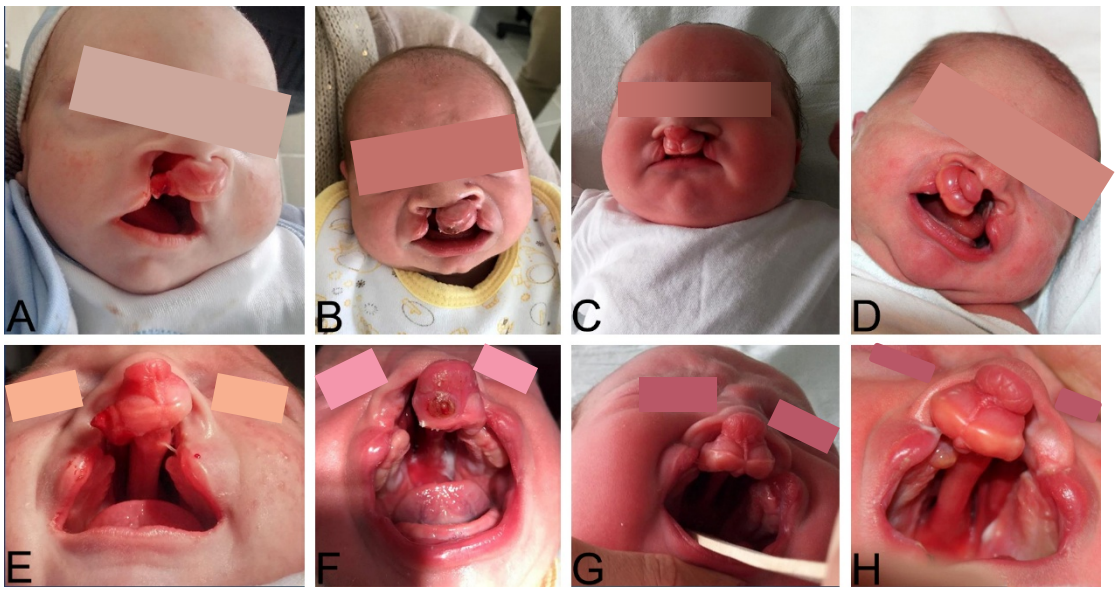

Fig. 2 – Clinical picture of bilateral cleft lip and palate (BCLP) in newborns: extraoral (A, B, C, and D) and intraoral (E, F, G, and H) presentation of BCLP in one-day-old newborns.

number of patients in whom the premaxilla is further protruded. The frequency of this subgroup of bilateral clefts is 4% of all orofacial clefts on average [3]. BCLP can be non-syndromic or part of over 400 described syndromes [4, 5]. The therapy represents a challenge, both from an aesthetic and functional perspective. It begins from the very birth of a person with a cleft and lasts for a long time. The necessity of starting therapy from the very beginning of life is justified because, otherwise, the risk of morbidity and mortality increases [6]. Surgical reparation of the lip is one of the priorities, and besides improving functionality, the goal is to achieve a more aesthetically acceptable appearance of the patient. The possibility of performing a surgical intervention and achieving good aesthetic results largely depends on the clinical picture of the patient; therefore, presurgical orthopedic care for the baby is extremely important. Despite the existence of a large number of treatment protocols, the ideal protocol has not yet been adopted either on the European or global level, although the difficulties caused by this type of cleft are significant. This paper provides an overview of different treatment protocols for infants with BCLP worldwide, as well as the treatment protocol using the RBJ (Radojičić Božidar and Julija – authors of the device) stimulator that is implemented in our country.

Bilateral cleft lip and palate

BCLP are considered the most severe clinical form of cleft lip and palate [1] and a reflection of severely violated morphological relationships of the split upper jaw. The entire upper jaw is divided into three parts so that the frontonasal process, which carries the premaxilla and the two maxillary extensions, is clearly visible. It seems that the frontonasal process protrudes out of the deepest parts of the nasal cavity, which makes the appearance of the newborn dramatic. Therefore, the procumbent and the rotated premaxilla is the stigma of this anomaly. There is also a significant increase in the width of the alar cartilage base, broadly separated lip segments, and a very short columella. Due to the disturbed physiological attachments of the muscles, the anomaly tends to worsen with growth and take on the feature of abnormality in untreated patients as they grow.

Functional, aesthetic, and therapeutic problems of patients with BCLP

The problems children with BCLP face manifest immediately after birth. Apart from other problems (malnutrition, infections, etc.), the two most significant ones are the inability to be fed through breastfeeding and significant aesthetic problems [7, 8]. Bearing in mind that they appear on the face, on the part of the body that is exposed to the views of other people and which cannot be hidden, the persons with this type of cleft also develop many psychological problems [9]. Severe aesthetic problems present at birth can become even more severe if the surgical reparation of the lip is not done perfectly. The therapy is highly complex, and the period prior to the first surgical intervention is crucial. During this time, growth is the most intense. Growth as a biological process does not differ from the growth of healthy individuals; therefore, any mistake in therapy can disrupt its course and have consequences for a lifetime.

The therapy depends on the development of the premaxilla, its size, shape, and rotation. Besides this, it also depends on the development of palatinal segments, the width of the cleft, both at the level of palatinal segments and alveolar ridges, the thickness, development, and shape of the vomer, the development of the prolabium, and the length of the columella. However, protrusion of the premaxilla remains the main stumbling block in the therapy of BCLP patients.

Protrusion of the premaxilla

The retraction of the premaxilla in its appropriate position to achieve a more functional and aesthetic reparation is the main goal in BCLP therapy [10, 11].

It is almost impossible to perform a surgical lip closure due to the considerable distance that exists between the prolabium and the muscular tissue of the lateral parts of the lip. Forced closure of these elements causes great tension in the sutures, which can lead to dehiscence. If dehiscence does not occur immediately, the consequences of such closure may become evident around the age of two. They are manifested by the vertical dropping of the premaxilla and its appearance outside the oral cavity at the level of the chin. Often, during this period, the incisors on the premaxilla have erupted, giving the entire clinical picture a very unaesthetic and undesirable appearance [3]. Such children are often teased about their "hot dog sticking out of their mouths" [12]. Correcting and returning the premaxilla to its normal position at this stage is even more difficult than in infancy. The reaction of the vomero-premaxillary suture (VPS) and the septomaxillary ligament is responsible for the increased sagittal growth of the premaxilla; thus, these two factors should always be taken into consideration from the beginning of therapy planning.

Vomero-premaxillary suture

The first evidence of the factor of protrusion was set by Veau (1934), who first correlated the excessive protrusion of the premaxilla with the excessive growth of the vomero-palatine structure resulting from the continuous formation of the bone or cartilage, but he did not histologically prove it [13]. The most convincing explanation of the factor contributing to the protrusion was established by Friede [14] in his study on the growth of VPS. It is very important for persons with BCLP because it is responsible for the rapid growth in the first postnatal months and is very fragile. Its activity is often referred to as "excessive growth" in both senses: the growth is greater than in people without a cleft, and it coincides with the protrusion of the premaxillary segment. It is a strategically important suture, as it is the only suture that can move the premaxilla to grow forward. Based on histological and X-ray analyses, he indicates the appearance of the secondary cartilage in the posterior part of the VPS, which occurs in response to mechanical loading in the period of rapid growth after birth. The secondary cartilage appears on both sides of the suture.

The septomaxillary ligament

Besides the VPS, the septomaxillary ligament contributes to the increased protrusion of the premaxilla. At the level of the VPS, this ligament connects the premaxilla to the nasal septum. At this level, a new bone is formed as a result of the tension appearing with the growth of the nasal septum and "carrying the premaxilla forward" [15]. The accelerated formation of the new bone requires the urgency of orthopedic therapy. Around the age of three, the newly formed bone, together with the surrounding bony structures and reduced tissue elasticity, compromises the success of therapeutic intervention. Verwoerd and Verwoerd-Verhoef [16] also point out the importance of caution regarding vomeral structures during therapy, highlighting their significance in the context of surgery. The part of the vomer in front of the premaxillary-vomeral suture is very fragile, and its damage can lead to irreversible consequences. For these reasons, the vomer osteotomy performed in patients with a protruded premaxilla should be done strictly distally from the VPS. In addition, with growth, there is a progressive ossification of the cartilaginous septum, extending to the perpendicular plate towards the alas of the vomer in the cranio-caudal direction. The loss of cartilage or damage to the vomer can occur as a result of surgical intervention performed between the vomer and the cartilage, which can lead to growth disturbances and deformities of the midface.

Age

It is extremely important to carry out the appropriate therapy at the very beginning of life because each mistake has consequences that cannot be corrected by any therapy.

Whether to operate on an infant or to initiate early orthodontic therapy is the question that continually raises a debate. The surgical intervention of connecting the lips in children with a highly protruding premaxilla has its specific risks. An incorrect therapy at the very beginning of life in a person with BCLP can have consequences, and the first signs of the wrong treatment can be visible at about the age of two [11]. First of all, they are related to malocclusions of dentofacial nature that can occur due to poorly performed surgical interventions of protruded premaxilla and vertically lowered premaxilla [17], or retrusion of the premaxilla and the entire middle floor of the face [13, 18]. A potential necrosis of the premaxilla is also possible. The significance of starting presurgical orthopedic therapy as early as possible is linked to the fact that the level of circulating estrogen, which the baby inherited from the mother, drastically decreases after the third month [19], as well as the contractility of the VPS [20]. After the eighth month, the contractility of the cartilaginous nasal structures is also reduced [21].

The shape and the size of the premaxilla

El-Kassaby et al. [22] study suggest a new descriptive classification of BCLP based on the characteristics of the premaxilla and indicates the dependence of the choice of therapy and the size of the premaxilla. Namely, the size of the premaxilla influences the results of both orthodontic and surgical therapies. Therefore, not all BCLP cases can be pooled under the same category or treated following the same rules, nor can we expect the same outcomes. All BCLPs were divided into the P group (protrusion of the premaxilla, longer prolabium) and the R group (resilient premaxilla, short prolabium). Small premaxilla occurs in people with a badly developed nasal septum, but the relationship of the premaxilla and the vomer is much more

flexible and better responds to orthodontic therapy (a more significant reduction in anteroposterior relationships of the premaxilla) and surgical therapy compared to those with the protruding premaxilla.

Therapy

Highly complex therapy starts at an early age and involves both presurgical orthopedic therapy and surgical therapy. These two are not mutually exclusive but rather interdependent.

Surgical therapy

In the past, the therapy for people with BCLP was solely focused on surgical lip reparation. As a consequence, a scope of different surgical techniques was developed, ranging from the osteotomy of the premaxilla, as the most radical and harmful method [18], to various surgical techniques. Some of these methods had an adverse effect on the development of the middle part of the face, which was indicated by orthodontists [23, 24]. The general impression was that after surgical procedures, patients resembled each other – they had a short columella, a stiff nasal tip, and widened nostrils. Better aesthetic results were achieved in the late 1980s. The study by McComb and Coghlan [25] showed that early neonatal surgery did not harm nasal growth, as previously thought. Consequently, the first operations, including nasal interventions, led to better aesthetic results.

Afterward, new surgical techniques were developed, aiming to improve the appearance of the nose while correcting the lip deformity, and the "columella in the nose" became a new approach in the surgical treatment of newborns with BCLP [26, 27].

Modern surgical therapy involves a large number of surgical modifications that vary among cleft centers.

Presurgical orthopedic therapy

Early on, it was realized that it was necessary to perform a retrusion of the premaxilla between previously aligned maxillary segments so as to create preconditions for good surgical therapy results, including a stable upper alveolar arch and the absence of oronasal fistulas.

Different presurgical procedures such as extraoral traction, oral pinning, premaxillary setback, and premaxillary excision [28] have been described. However, in all of the described methods, the impact on the development of maxillary growth was possible in the sense that they could have a negative impact. In the literature, two main techniques for presurgical manipulation of the premaxilla have been described – passive and active. Passive plates did not affect the reduction of the cleft size between the alveolar ridges and palatinal segments, and the retraction of the premaxilla was performed by an external force that was not part of the appliance.

Active intraoral appliances were described by Reisberg et al. [29]. Some authors directed the growth of the premaxilla in the downward and backward direction by application of force on the premaxilla [30–32]. However, some opposed presurgical orthopedic therapy. Millard et al. [33] suggest that any application of force could have a restriction impact on the growth of the premaxilla. Hotz et al. [34] thought that passive intraoral appliances should direct skeletal growth in the desired direction or even stimulate growth, which Weil [35] and Nolst et al. [36] confirmed in their studies. Both active and passive appliances incorporated into the lateral segments provide a stabilizing effect on them. All the devices described so far had different types of fixations that helped retain the apparatus in the mouth of the newborn. On the other hand, they caused certain complications to the development of the upper jaw. These complications included appliances where insertion of trans-premaxillary pins [30, 31] was performed, but this raised the probability of damaging the developing tooth buds and was not suitable for use in older age groups apart from being technique sensitive.

Georgiade technique

This technique involves the application of an active dentofacial orthopedic appliance [31]. A manually produced appliance based on the upper jaw impression is inserted into the baby's mouth under general anesthesia. Acrylic plates are secured (screwed) to the palatinal segments. A wire loop is passed through the premaxilla's neck at the level of the VPS. One elastic chain on each side connects the transmaxillary wire, goes to the posterior part of the plate, passes underneath, and then returns to the front edges of the appliance on each side. Parents turn the screw positioned in the middle of the appliance, increasing the distance between the palatinal segments only in the anterior, alveolar width of the cleft. The orthodontist reduces the length of the elastic chain, retracting the premaxilla in this way. It takes six to eight weeks to align the premaxilla between the separated palatinal segments.

The Latham technique

The Latham appliance was the most effective in correcting the premaxilla position in the anteroposterior direction. However, the movement was mostly retroinclination rather than retroposition. The teeth became retroinclined, and the vomer was slightly curved. This appliance also corrected premaxillary rotation but had a small impact on the vertical position [15]. Nasoalveolar molding (NAM) is a newer presurgical orthopedic appliance in the development of presurgical orthopedic therapy. It arose from the need to correct nasal cartilage deformity and columella tissue deficiency before a surgical lip closure, thus eliminating the need for later surgical columella lengthening. Mazaheri et al. [37] emphasize that the main advantage of NAM is the possibility of performing lip and nasal surgery in a single procedure, by which better aesthetic results are achieved.

Grayson's nasoalveolar molding technique

Grayson et al. [38] started their technique in 1990. The appliance first performs alveolar molding, reducing the alve-

olar cleft to less than 5 mm. Nasal molding is then conducted through a nasal molding element added to the basic palatal passive plate. Stretching of the short columella is done using two acrylic nasal stents on the acrylic extension attached to the passive plate. For further columella stretching, a horizontal soft strip is used, while a vertical strip is placed from the prolabium to the appliance to provide a counterforce. At the same time, the retraction of the premaxilla is gradually carried out through the serial application of the strips going to the bottom of the cheeks or the lips. The strips are changed daily by the parents. The appliance must be adjusted every week in order to modify the alveolar molding plate.

Figueroa's nasoalveolar molding technique

The appliance described by Bennun and Figueroa [39] does not use nasal extension strips. The acrylic intraoral appliance rests loosely in the mouth. During sucking and swallowing, lingual movements transmit force *via* a flexible spring to the nasal extension with silicone stops. All techniques with external traction applied directed pressure to the lower basal part of the premaxilla, causing lingual inclination and curving of the vomer. Furthermore, it was difficult to centralize a severely rotated premaxilla with passive plates and external traction.

Liou's nasoalveolar molding technique

This technique uses dental adhesive to provide retention of the acrylic plate to the maxilla. The additional nasal component consists of a curved steel wire with balls made of soft acrylic, which appear on both sides of the dental plates. They are positioned in the nostrils and make movements forward and upward. The dental plate molds the maxillary segments, while the added acrylic balls positioned under the surface of the nasal cartilages mold the nasal cartilages. The technique also uses strips over the lips in order to retract the premaxilla, bring the alar bases closer together, and reduce the alveolar cleft area in this way. That contributes to a good nose configuration. The columella is lengthened by simultaneously pushing the premaxilla backward and the nose tip forward [40]. Numerous centers have replaced their protocols in the treatment of patients with BCLP and applied this method [12, 41]. However, in order to adopt this method, further research is needed to confirm its long-term positive effects [7]. The latest method applied in patients with BCLP involves NAM, followed by simultaneous surgical lip reparation and primary rhinoplasty. That results in a columella of approximately normal length until young adulthood. However, secondary nose corrections will be necessary for one-third of the operated patients since larger widths of all nasal characteristics are present when compared to individuals without a cleft [42].

RBJ stimulator

Presurgical orthopedic therapy *via* RBJ stimulator is one of the attempts to solve the problem of a severe clinical picture of a newborn with BCLP in the best possible way [43]. The aim is to bring the premaxilla and the lateral, palatinal segments to a most proper relationship prior to the first surgical intervention, thus creating the conditions for achieving a high success rate of future surgery. This therapy is based on the biological concept and the aspect of individuality. It starts from the earliest age, at the very beginning of life, by manufacturing an obturator and a stimulator. The procedure of obtaining a two-phase impression – anatomical (Figure 3A–C) and functional (Figures 3D–F and 4A), is of crucial importance for the precision and quality of the plaster model (Figure 4B), based on which the RBJ stimulator is manually produced (Figure 4C). Great knowledge of the embryonic development of the orofacial region, as well as the "attentiveness" to the craniofacial region in persons with BCLP, the recognition of the type of growth and the experience of doctors are the key to cleft care in persons with BCLP. An orthodontist should guide and direct the growth from birth to adolescence according to the very characteristics of the growth and the development of the cleft. Although they are

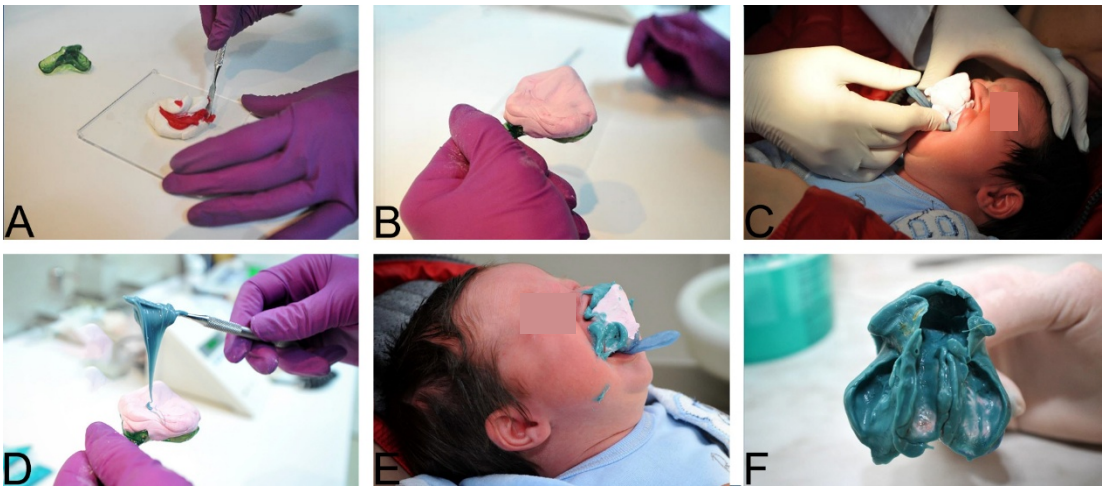

Fig. 3 – A two-phase process of obtaining the impression of the upper jaw in a newborn with bilateral cleft lip and palate: phases of obtaining the anatomical (alginate) impression (A, B and C); phases of obtaining the functional (corrective) impression (D, E and F).

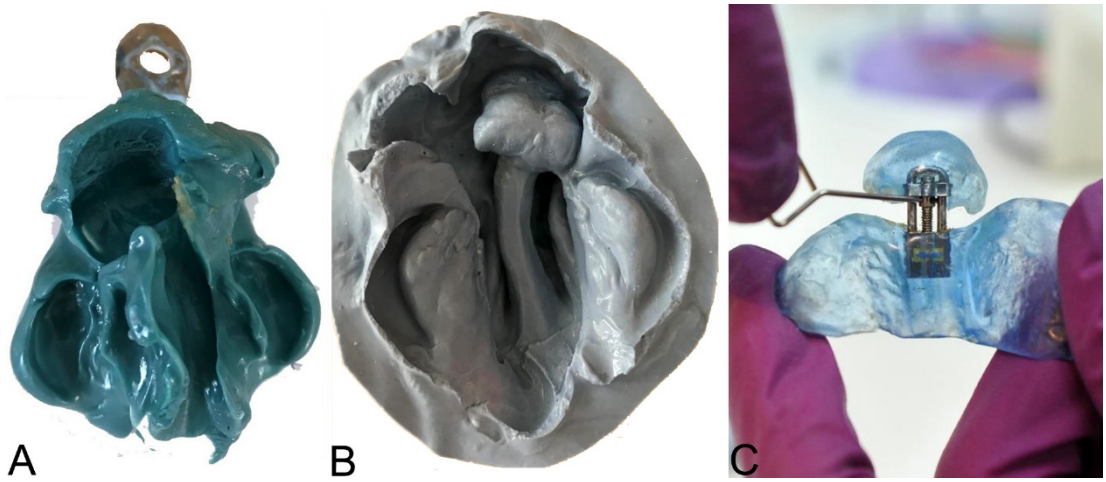

Fig. 4 – Impression (A), plaster model (B), and RBJ stimulator (C) of one-day-old newborn with bilateral cleft lip and palate. RBJ – Radojičić Božidar and Julija

similarly classified, they are not the same. Therefore, each case requires differential diagnosis and specific treatment planning. What may be the chosen treatment for one patient may be different for another, even if they have the same cleft type. Our results and attitudes regarding tracking and directing the growth are in line with Meazzini et al. [7].

Among the multitude of the described apparatuses, RBJ has its own place. For the stimulator to show its positive effect, it must have a well-insured base. Only the stimulator that intimately adheres to the upper jaw mucosa can be effective. Each shaking in the mouth, securing with the denture adhesive, various elastic strips, caps, etc., lead to the failure of the presurgical therapy [44]. A good base is established only by a precisely taken impression, which is, in fact, a key factor. A well-secured base (the secondary palate completely covered with acrylate: alveolar edges, palatine extensions with maxillary tubers included, up to the border with soft palate) is an ideal pressure site, which allows a smaller segment, premaxilla, to direct the path backward between the laterally set palatine segments using the orthodontic screw. It is best to start the therapy immediately after birth because, even after eight months, premaxillary-vomerine juncture, as well as maxillary segments, become stiff [21]. A rigid bond of acrylate that covers the premaxilla and the activation of the screw lead the premaxilla backward, affecting the premaxillary-vomerine juncture but also the nasal septum, not allowing it to continue to grow forward.

The question that arises is about the significance of the application of therapy with these devices. The stimulator does not use strong force for a short period of two to three weeks as Latham appliance, thus retracting the anterior nasal spine in conjunction with the alveolar region of the premaxilla [15].

The strategy for the development of the stimulator is in agreement with the studies of Friede and Morgan [13], who argue that an inadequate mechanical load at the VPS level can provoke border cells on both sides of the suture to form the secondary cartilage, which is responsible for excessive growth of the premaxilla, both in the sagittal and vertical directions. The most common type of such mechanical load is the inadequate extraoral or intraoral fixation of the appliance. The stimulator, as indicated, has excellent retention in the mouth of the newborn, so this kind of unpleasant pressure on the premaxilla is not present. The pressure acting on the above-mentioned structure of the damaged jaw and the force acting on the premaxilla is controlled by the stimulator design and by the speed and the number of rotations of the screw. The reduction of the protrusion of the premaxilla is performed by the stimulator through bone contraction by gradual compression, which is closely related to the mechanism of operation of the device by Liou et al. [40], and there is no resorption of the posterior part of the VPS. After the early orthodontic treatment with a stimulator and by achieving retrusion of the premaxilla, the vertical lowering of the premaxilla is avoided, which occurs if the lip surgery is performed without prior bringing the premaxilla in a corresponding sagittal position in relation to the lateral extensions. In that case, first, the resorption occurs at the posterior border (oral surface) of the vomeropalatine suture, and only then is the secondary cartilage that leads the premaxilla vertically downward created [12]. In addition, the stimulator limits the mobility of the premaxillary segment, which is also stated as one of the favorable factors in the induction of secondary cartilage – intermittent pressure. It occurs due to the absence of normal musculature, the absence of connective tissue over the split [24], and the presence of anterior pressure by the lower lips and the tongue [45]. The stimulator prevents increased mobility not only by rigidly securing the segment of the mentioned complex but also by disabling the pressure from the tongue and the lower lip. The great advantage of the RBJ stimulator in the sense of the absence of an extraoral fixation in its design is that there are no complications such as skin injuries caused by the adhesion tape, the stuffy appearance with the headband, or the patient's and parent's compliance. Furthermore, the results are not highly predictable [46].

Early presurgical therapy through stimulators is in agreement with the studies that advocate the thesis that, apart from a great knowledge of embryonic development, the

knowledge of craniofacial growth is also very important for the therapy [47]. Boo-Chai [47] stated that if the surgery has not been performed on the jaw, it will grow normally (normal relationships with other parts of the face), which means that the presence of the cleft is not the factor in the inhibition of the growth process. These findings are also consistent with Capelozza Filho et al. [48], who state that in the absence of surgery, the jaw will be able to reach the normal anteroposterior dimension. In addition, the findings are consistent with the recent study by Shetye and Evans [49] including the significance of the protrusion of point A in comparison to the control sample with the mandibular retrusion, and a much wider cranial base angle. All of this confirms our view that if the growth potential exists, it is possible to influence the life of the newborn with BCLP in the earliest days with appropriate devices, such as the stimulator. The therapy through the RBJ stimulator is conceived so that it acts safely on the VPS. The VPS is very sensitive, and poor therapy can cause its damage (severe retrusion of the middle floor of the face). Its growth can be influenced, but one has to be cautious. The therapy with the stimulator avoids complications that occur later in childhood. Nevertheless, by comparing them with the results of Oosterkamp et al. [50], in whose research a considerable retrusion of the premaxilla occurred, the retrusion through the RBJ stimulator was twice as high. The total value of the retrusion of the premaxilla was statistically significantly reduced in the course of six months due to the activation of the orthodontic screw and the construction of a stimulator that prevented the unrestricted growth of the septum and premaxilla from further sagittal "rampage". In order to avoid all the complications that can occur during the growth of a person with BCLP, it is necessary to orthodontically make an effect on the upper jaw damaged by a cleft at the earliest possible age, i.e., immediately after birth. Therefore, the results of this study show that it is possible to achieve good clinical effects in newborns with BCLP with the stimulator, although McNeil [51] proved in 1950 that it is difficult to design an apparatus that can simultaneously close the cleft and reposition the premaxilla. The application of the stimulator is considered irreplaceable in the period immediately after birth (it provides nutrition), and with all its characteristics, which primarily originate in its design without extraoral fixation, it makes excellent preparation for future successful surgical interventions, corrects severe aesthetic problems without harming subsequent growth, and reduces the need for implementing other treatment techniques (e.g., bone grafting) during growth.

If we compare the effects of the RBJ stimulator with the current NAM, its main goal is columella elongation and improvement of the aesthetic results of lip and nose surgery. A skeletal effect on the palatinal segments and protrusion of the premaxilla has not been demonstrated, except that it does not seem to have negative effects on skeletal development after craniofacial growth has been completed compared to the group of patients treated without NAM [52]. Radojičić [53] showcases the positive effects of the RBJ stimulator in a study based on a three-dimensional analysis of the effects of the early orthodontic therapy *via* stimulator of a specific construct by taking impressions, conventionally, of 50 newborns with different types of clefts (unilateral cleft lip and palate, BCLP, cleft palate) immediately after birth, and then continuously each month. Active stimulator effects are based on the application of basic biomechanical principles adapted to the individualized cleft anatomy [19, 36], excellent retention of the stimulator achieved without extraoral or intraoral fixation [20] (and thus avoiding harming of subsequent maxilla growth [54–58], effects from the first hours of life), maximal use of biological potentials – suppleness of cartilaginous structure [59], and the elimination of the complications of the ossified protruding premaxilla [21]. Until the conventional method of making RBJ stimulators is replaced by intraoral 3D scanning and the manufacturing of stimulators or feeding appliances by Computer-Aided Design/Computer-Aided Manufacturing (CAD/CAM) technique, we consider that this method of manufacturing greatly influences the achievement of excellent presurgical therapy because the strategy of treatment and the design of our stimulator are identical to 3D CAD/CAM stimulators [53].

Conclusion

The presented overview of the main techniques for conducting presurgical orthopedic therapy in patients with BCLP provides the basic characteristics of their actions, positive effects, as well as drawbacks that require further modifications to achieve even better results. Thus, a definitive therapeutic treatment that could be recommended as the treatment of choice has not been selected yet. The RBJ stimulator applied in Serbia as a unique therapeutic solution is particularly highlighted. Its advantages compared to the other therapeutic techniques include the following characteristics: it is the only appliance whose palatal plate has an active function; by moving the palatinal segments, the entire zygomatic complex is moved through the sutures they are connected to, and by retracting the protruded premaxilla, the nasal complex is moved, thus influencing the orthopedics of the entire face; using the active effect skeletal base, balance is achieved, and the need for bone grafting is eliminated. Owing to these characteristics, the drawbacks of other described appliances have been overcome, and the surgical results of lip reparation after the application of orthopedic therapy using the RBJ stimulator in newborns with BCLP are of high aesthetic and functional quality. Therefore, considering all the mentioned, the application of the RBJ stimulator can be suggested as the recommended form of presurgical orthopedic therapy in patients with BCLP.

Conflict of interest

The authors declare no conflict of interest.

R E F E R E N C E S

1. *Radojicic J*, *Trifunovic B*, *Cutovic T*, *Radojicic A*. Presurgical Orthopedic Treatment of a 3-Year-Old Child with Unoperated Complete Bilateral Cleft Lip and Palate and Protruding Premaxilla. Cleft Palate Craniofac J 2023; 60(5): 627–34.
2. *Mahmood R*, *Flood T*, *Robinson S*, *Al-Gholmy M*. Early orthopedic retraction of the premaxilla in bilateral complete cleft lip and palate: an innovative approach to a difficult problem. Cleft Palate Craniofac J 2016; 53(3): 363–7.
3. *Latief BS*, *Lekkas C*, *Kuijpers MA*. Maxillary arch width in unoperated adult bilateral cleft lip and alveolus and complete bilateral cleft lip and palate. Orthod Craniofac Res 2010; 13(2): 82–8.
4. *Radojičić J*. The application of stimulator in the treatment of cleft lip and palate in Goldenhar syndrome, trisomy 13 and lobar holoprosencephaly with a median cleft lip. FU Med Biol 2018; 20(1): 24–9.
5. *Radojicic J*, *Tanic T*, *Pesic Z*, *Jovic N*, *Cutovic T*, *Filipovic G*. Rare disease: lobar holoprosencephaly with a median cleft lip-case report. Cleft Palate Craniofac J 2016; 53(1): 109–17.
6. *Shkoukani MA*, *Chen M*, *Vong A*. Cleft lip - a comprehensive review. Front Pediatr 2013; 1: 53.
7. *Meazzini MC*, *Rossetti G*, *Morabito A*, *Garattini G*, *Brusati R*. Photometric evaluation of bilateral cleft lip and palate patients after primary columella lengthening. Cleft Palate Craniofac J 2010; 47(1): 58–65.
8. *Vyas RM, Kim DC, Padwa BL, Mulliken JB*. Primary Premaxillary setback and repair of bilateral complete cleft lip: indications, technique, and outcomes. Cleft Palate Craniofac J 2016; 53(3): 302–8.
9. *Persson M*. Cleft lip and palate in adolescence identifying variables relating to psychosocial concerns [Ph.D. Thesis]. Lund, Sweden: Lund University Faculty of Medicine; 2007.
10. *ECPCA*. ECPCA Craniofacial Orthodontics Course 2022-2023. Craniofacial Anomalies and Cleft Lip and Palate: Clinical Principles and Orthodontics Management [Internet]. Milan: ECPCA; 2023 [updated 2023 June 24; cited 2023 June 26]. Available from: https://ecpcainternationalcourse2022-2023.it/
11. *Meazzini MC*, *Cohen N*, *Autelitano L*, *Radojicic J*. Non-surgical treatment of vertical excess of the premaxilla in growing bilateral cleft lip and palate patients. Int J Oral Maxillofac Surg 2021; 50(12): 1576–82.
12. *Meazzini MC*, *Brusati R*, *Bozzetti A*, *Mazzoleni F*, *Felisati G*, *Garattini G*, et al. Craniofacial Anomalies: Surgical-orthodontic Management. Bologna: Edizioni Martina; 2011.
13. *Friede H*, *Morgan P*. Growth of the vomero-premaxillary suture in children with bilateral cleft lip and palate. A histological and roentgencephalometric study. Scand J Plast Reconstr Surg 1976; 10(1): 45–55.
14. *Friede H*. Histology of the premaxillary=vomerine suture in a bilateral cleft case. Cleft Palate J 1973; 10: 14–22.
15. *Latham RA*. Development and structure of the premaxillary deformity in bilateral cleft lip and palate. Br J Plast Surg 1973; 26(1): 1–11.
16. *Verwoerd CD*, *Verwoerd-Verhoef HL*. Rhinosurgery in children: developmental and surgical aspects of the growing nose. GMS Curr Top Otorhinolaryngol Head Neck Surg 2010; 9: Doc05.
17. *Meazzini M*, *Lematti L*, *Mazzoleni F*, *Rabbiosi D*, *Bozzetti A*, *Brusati R*. Vertical excess of the premaxilla in bilateral cleft lip and palate patients: a protocol for treatment. J Craniofac Surg 2010; 21(2): 499–502.
18. *Millard DR*. Cleft Craft: The Evolution of its Surgery. II. Bilateral and Rare Deformities. Volume 2. Boston: Little Brown and Co; 1977. p. 922.
19. *Semb G*, *Shaw WC*. Facial growth related to surgical methods. In: *Berkowitz S,* editor. Cleft lip palate: Diagnosis and Management. 3rd ed. Heidelberg, Berlin: Springer; 2013. p. 325–43.
20. *Georgiade NG*, *Latham RA*. Maxillary arch alignment in the bilateral cleft lip and palate infant, using pinned coaxial screw appliance. Plast Reconstr Surg 1975; 56(1): 52–60.
21. *Mulliken JB*, *Wu JK*, *Padwa BL*. Repair of bilateral cleft lip: review, revisions, and reflections. J Craniofac Surg 2003; 14(5): 609–20.
22. *El-Kassaby MA*, *Abdelrahman NI*, *Abbass IT*. Premaxillary characteristics in complete bilateral cleft lip and palate: A predictor for treatment outcome. Ann Maxillofac Surg 2013; 3(1): 11–9.
23. *Heidbuchel KL*, *Kuijpers-Jagtman AM*, *Freihofer HP*. An orthodontic and cephalometric study on the combined surgical-orthodontic approach of the protruded premaxilla in bilateral clefts. J Craniomaxillofac Surg 1993; 21(2): 60–6.
24. *Pruzansky S*. The growth of the premaxillary-vomerine complex in complete bilateral cleft lip and palate. Tandlaegebladet 1971; 75(12): 1157–69.
25. *McComb HK*, *Coghlan BA*. Primary repair of the unilateral cleft lip nose: completion of a longitudinal study. Cleft Palate Craniofac J 1996; 33(1): 23–30; discussion 30–1.
26. *Mulliken JB*. Bilateral complete cleft lip and nasal deformity: an anthropometric analysis of staged to synchronous repair. Plast Reconstr Surg 1995; 96(1): 9–23; discussion 24–6.
27. *Trott JA*, *Mohan N*. A preliminary report on one stage open tip rhinoplasty at the time of lip repair in bilateral cleft lip and palate: the Alor Setar experience. Br J Plast Surg 1993; 46(3): 215–22.
28. *Suri S*, *Tompson BD*. A modified muscle-activated maxillary orthopedic appliance for presurgical nasoalveolar molding in infants with unilateral cleft lip and palate. Cleft Palate Craniofac J 2004; 41(3): 225–9.
29. *Reisberg DJ*, *Figueroa AA*, *Gold HO*. An intraoral appliance for management of the protrusive premaxilla in bilateral cleft lip. Cleft Palate J 1988; 25(1): 53–7.
30. *Bitter K*. Latham's appliance for presurgical repositioning of the protruded premaxilla in bilateral cleft lip and palate. J Craniomaxillofac Surg 1992; 20(3): 99–110.
31. *Georgiade NG*, *Mason R*, *Riefkohl R*, *Georgiade G*, *Barwick W*. Preoperative positioning of the protruding premaxilla in the bilateral cleft lip patient. Plast Reconstr Surg 1989; 83(1): 32–40.
32. *Papay FA*, *Morales L*, *Motoki DS*, *Yamashiro DK*. Presurgical orthopedic premaxillary alignment in cleft lip and palate reconstruction. Cleft Palate Craniofac J 1994; 31(6): 494–7.
33. *Millard DR*, *Latham R*, *Huifen X*, *Spiro S*, *Morovic C*. Cleft lip and palate treated by presurgical orthopedics, gingivoperiosteoplasty, and lip adhesion (POPLA) compared with previous lip adhesion method: a preliminary study of serial dental casts. Plast Reconstr Surg 1999; 103(6): 1630–44.
34. *Hotz M*, *Perko M*, *Gnoinski W*. Early orthopaedic stabilization of the premaxilla in complete bilateral cleft lip and palate in combination with the Celesnik lip repair. Scand J Plast Reconstr Surg Hand Surg 1987; 21(1): 45–51.
35. *Weil J*. Orthopaedic growth guidance and stimulation for patients with cleft lip and palate. Scand J Plast Reconstr Surg Hand Surg 1987; 21(1): 57–63.
36. *Nolst TG*, *Weil J*, *de Roos P*. Observation: a comment on "A discussion of presurgical orthodontics in patients with clefts". Cleft Palate J 1990; 27(4): 419–23: discussion 423–4.
37. *Mazaheri M*, *Harding RL*, *Cooper JA*, *Meier JA*, *Jones TS*. Changes in arch form and dimensions of cleft patients. Am J Orthod 1971; 60(1): 19–32.

38. *Grayson BH*, *Santiago PE*, *Brecht LE*, *Cutting CB*. Presurgical nasoalveolar molding in infants with cleft lip and palate. Cleft Palate Craniofac J 1999; 36(6): 486–98.
39. *Bennun RD, Figueroa AA*. Dynamic presurgical nasal remodeling in patients with unilateral and bilateral cleft lip and palate: modification to the original technique. Cleft Palate Craniofac J 2006; 43(6): 639–48.
40. *Liou EJ*, *Chen PK*, *Huang CS*, *Chen YR*. Orthopedic intrusion of premaxilla with distraction devices before alveolar bone grafting in patients with bilateral cleft lip and palate. Plast Reconstr Surg 2004; 113(3): 818–26.
41. *Meazzini MC*, *Chiavenna C*, *Autelitano L*, *Garattini G*, *Brusati R*. Photometric Evaluation in Adolescence of Patients With Bilateral Cleft Lip and Palate Treated With Nasoalveolar Molding and Primary Columella Lengthening. Cleft Palate Craniofac J 2018; 55(4): 568–73.
42. *Meazzini MC*, *Parravicini F*, *Donati V*, *Brusati R*, *Biglioli F*, *Autelitano L*. Photometric Evaluation of Adult Patients With Bilateral Cleft Lip and Palate Treated With Nasoalveolar Molding and Primary Columella Lengthening. Cleft Palate Craniofac J 2022; 59(7): 852–8.
43. *Radojičić J*, *Tanić T*, *Jović N*, *Cutović T*, *Papadopoulos K*. Presurgical orthodontic treatment of patients with complete bilateral cleft lip and palate. Vojnosanitet Pregl 2014; 71(7): 693–9.
44. *Nemes B*, *Fábián G*, *Nagy K*. Management of prominent premaxilla in bilateral cleft lip and alveolus. Cleft Palate Craniofac J 2013; 50(6): 744–6.
45. *Glass D*. The early management of bilateral cleft of lip and palate. Br J Plastic Surg 1970; 23(2): 130–41.
46. *Kiya K*, *Oyama T*, *Sone Y*, *Ishii N*, *Hosokawa K*. A novel active intraoral appliance for presurgical orthopaedic treatment in patients with complete bilateral cleft lip and palate. J Plast Reconstr Aesthet Surg 2015; 68(5): 632–7.
47. *Boo-Chai K*. The unoperated adult bilateral cleft of the lip and palate. Br J Plast Surg 1971; 24(3): 250–7.
48. *Capelozza Filho L*, *Normando AD*, *da Silva Filho OG*. Isolated influences of lip and palate surgery on facial growth: comparison of operated and unoperated male adults with UCLP. Cleft Palate Craniofac J 1996; 33(1): 51–6.
49. *Shetye PR*, *Evans CA*. Midfacial morphology in adult unoperated complete unilateral cleft lip and palate patients. Angle Orthod 2006; 76(5): 810–6.
50. *Oosterkamp BC*, *van Oort RP*, *Dijkstra PU*, *Stellingsma K*, *Bierman MW*, *de Bont LG*. Effect of an intraoral retrusion plate on maxillary arch dimensions in complete bilateral cleft lip and palate patients. Cleft Palate Craniofac J 2005; 42(3): 239–44.
51. *McNeil CK*. Orthodontic procedures in the treament of congenital cleft palate. Dent Rec (London) 1950; 70(5): 126–32.
52. *Meazzini MC, Parravicini F, Cohen N, Rossetti G, Autelitano L*. Nasoalveolar molding and skeletal development in patients with bilateral cleft lip and palate: A retrospective cephalometric study at the completion of growth. J Craniomaxillofac Surg 2022; 50(5): 400–5.
53. *Radojicic J*. Cleft Care: Intraoral 3D Scanning. Cleft Palate Craniofac J. 2018: 55(9): 1330.
54. *Berkowitz SA*. Comparison of treatment results in complete bilateral cleft lip and palate using a conservative approach versus Millard-Latham PSOT procedure. Semin Orthop 1996; 2(3): 169–84.
55. *Berkowitz S*. Apoplectic or apocalyptic? J Oral Maxillofac Surg 2001; 59(10): 1252–3. Erratum in: J Oral Maxillofac Surg 2001; 59(12): 1516.
56. *Berkowitz S*. Primary repair of cleft lip and nasal deformity. Plast Reconstr Surg 2002; 109(6): 2158–61.
57. *Henkel KO*, *Gundlach KK*. Analysis of primary gingivoperiosteoplasty in alveolar cleft repair. Part I: facial growth. J Craniomaxillofac Surg 1997; 25(5): 266–9.
58. *Uzel A*, *Alparslan ZN*. Long-term effects of presurgical infant orthopedics in patients with cleft lip and palate: a systematic review. Cleft Palate Craniofac J 2011; 48(5): 587–95.
59. *Scott JH*. The cartilage of the nasal septum. Br Dent J 1953; 95: 37–40.

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