CASE REPORT

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Presurgical orthodontic treatment of patients with complete bilateral cleft lip and palate

Prehirurško ortodontsko lečenje bolesnika sa potpunim bilateralnim rascepom usne i nepca

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Abstract

Introduction. Cleft lips and palates are the most common congenital orofacial anomaly. This type of clefts is the most severe from the orthodontic-surgical therapy aspect. **Case report.** A female newborn with a complete cleft of the primary and the secondary palate was admitted to the clinic, where a multiple-role orthodontic device was specially designed and applied to primarily manage the closure of the existing cleft and help to improve the suckling ability of the baby. Besides the fact that it allows breastfeeding, it has a significant orthodontic effect, too. **Conclusion.** Specificity of this device is the lack of extraoral fixation. What can easily be observed is a progressive reduction of the cleft between the separated segments and the premaxilla retrusion. It, thus, allows the creation of much better conditions for further surgical management of the said defect.

Key words:
cleft lip; cleft palate; infant newborn; palatal obturators.

Apstrakt

Uvod. Rascepi usne i nepca su najčešće kongenitalne orofacijalne anomalije. Ovaj tip rascepa je najteži sa aspekta ortodontsko-hirurške terapije. **Prikaz slučaja.** Žensko novorođenče sa kompletnim rascepom primarnog i sekundarnog palatuma primljeno je na kliniku gde je izrađen višenamenski ortodontski aparat specifične konstrukcije koji omogućava ishranu, ali ima i značajan ortodontski efekt. **Zaključak.** Specifičnost ovog aparata je odsustvo ekstraoralne fiksacije, a zahvaljujući njemu dolazi do progresivne redukcije rascepa između segmenata, kao i retruzije premaksile. Na ovaj način stavaraju se daleko bolji uslovi za hiruršku terapiju koja sledi.

Ključne reči:
usna, rascep; nepce, rascep; novorođenče; opturatori; palatinalni.

Introduction

Cleft lips and palates are among the most common congenital orofacial anomalies. They occur in approximately 700 children born every day somewhere in the world (about 240, 000 newborns a year), and that number is ever increasing [1]. Its etiology has not been completely understood yet despite the fact that the heredity plays a crucial role in its occurrence. Clefts are inherited as a quasi-continuous genetic model with a distinct expressiveness. Cleft palate occurs in about one in 700 live births worldwide.

If we do not take oblique and transversal facial clefts into account, a complete bilateral cleft lip and palate are considered the most severe clinical form.

From the orthodontic-surgical therapy aspect, the clinical picture of this type of cleft is the most complex and severe, because the location of cleft segments is such that they are difficult to be surgically reponed in a way to allow good surgical restitution without the occurrence of postoperative complications such as development of the oronasal fistula. However, the primary problem arising immediately after birth of babies with such a severe deformity is feeding [2–4].

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It is thought that a large gap in children with this type of cleft causes a difficulty in creating an adequate negative pressure within the oral cavity due to the existence of a large oronasal communication. Such a communication prevents normal flow of fluid during breastfeeding and permits the entry of milk or water into the unprotected upper airways, what may result in some very dramatic situation [5]. Nasal regurgitation, an excessive air swallowing and milk aspiration can occur and may be followed by the episodes of choking, coughing, vomiting, difficulty in swallowing and cyanosis [4, 6]. Those babies, even if partially breastfed, are always tired and sleepy. All that can negatively impact the baby's growth [7–11]. The parents' stress when nursing their babies as well as their increased tension and worry because of their babies' insufficient milk intake should not be neglected since it has major effects on the baby's poor weight gain [12]. The mother-child relationship often changes as well [13].

The orthodontic aspect takes a special place in the treatment of cleft newborns. In those children born with bilateral cleft lip and palate, the premaxillary segment is very often protruded forward over the lateral palatinal segments and the lower jaw. The prolabium is very small in size with a hypoplastic muscular layer, and the muscle fibers of the orbicularis oris muscle are inserted near the alar base. A constant retraction not opposed by the lateral lip's muscles makes the diameter of the nose base wider than in the non-cleft cases. Columella is often short, or does not exist, so the nose is flat [14]. The first surgical intervention those patients need is lip adhesion, which could be performed in one or two phases. It depends on the premaxilla position and the quantity of the available tissue. The purpose of early orthodontic treatment is to minimize the need for an additional surgical treatment. Thus, a much better symmetry could be achieved, and visible scars may be minimized. In cases with the premaxilla protruded to a higher degree, one-phase surgical treatment helps the surgeon to reduce the distance between the premaxilla and the cleft lateral segments prior to the surgery.

The families with children born with cleft lip/cleft palate are exposed to stressful life events having social, financial, and mental implications to a much greater extent because clefts palate repair is performed at the time when their children reach the age of two [15].

The aim of this paper was to present the way a stimulator could be fabricated and applied to the baby with severe complete bilateral cleft of the primary and the secondary palate immediately after birth. In the first phase, it would be used as a passive palatal obturator to help the normalization of the baby's suckling ability, and, in the second phase, as an active obturator to allow the initial reposition of the separated segments.

Case report

A duly born female baby with the birth-weight of 2,200 grams and the length of 46 cm was diagnosed with severe and non-syndrome complete bilateral cleft of the primary and the secondary palate (Figure 1).

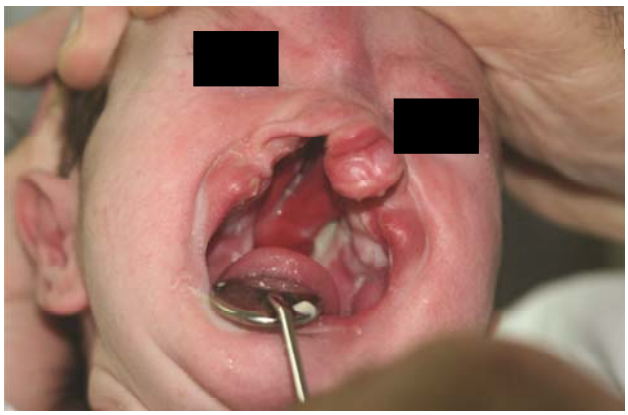

Fig. 1 – Intraoral appearance of the baby with complete bilateral cleft lip and palate.

A couple of hours after birth, the baby was referred to the clinic, and the initial step taken was to manage the problem and allow breastfeeding to baby by fabricating the first RBJ stimulator for closure of the communication between the nasal and the oral cavity.

The first stage in the fabrication of RBJ (Radojičić Božidar and Julija – the authors of the device) obturator is taking an anatomical impression of the upper jaw, which requires an exceptional professional expertise of orthodontists. Premedications given to reduce the stress in infants could provoke prolonged sedation in the postanesthesia period, so they may experience some functional disorders such as eating and sleeping problems even 15 days after the intervention. However, the endotracheal intubation has proved to be difficult in a ceratain number of such cases (4–7% of infants), and endotracheal tube itself covers a considerable space within the oral cavity, which makes taking a proper dental impression extremely complicated and affects both the precision of the obtained impression and the fabrication of an obturator.

The preliminary impression is anatomic, and is taken using an irreversible alginate impression material. The next stage is making an individual tray from this first impression in the dental laboratory, which is required for obtaining the second (corrective, functional) impression. It is very critical for the construction of the obturator because it should accurately show all the details of the baby's alveolar ridges (creases, frenulum). Therefore, impression mass consisting of addition silicones from the Group A featuring a high accuracy, a long-lasting dimensional stability, the hydrocompatibility, thixotropy and hydrophilicity should be used for such a purpose.

When compared to the standard impression technique, the procedure of taking corrective impressions required for the fabrication of a stimulator is specific in the ratio between the quantity of the activator and the quantity of the catalyst of addition silicone. Therefore, a special attention should be paid to that in this phase. The amount of the activator is twice the average amount used in the standard impression procedure, what provides more rapid bonding of the impression mass. The individual tray filled with such prepared impression mass is inserted into the baby's mouth exactly when the process of bonding starts (Figure 2). The baby's body

Fig. 2 – A day after birth, the baby's upper jaw impression was taken.

temperature accelerates the process even more, so the entire process takes about 40 seconds to complete, and the tray can be removed from the baby's mouth. However, this should be done with caution since an excessive amount of activator can have the opposite effect, in terms of activator's inability to bind to the catalyst of the impression mass.

When the individual tray with the impression mass is inserted into the baby's mouth, it is necessary to put the left hand on the baby's cheek to manually palpate the cheek muscles, i.e. to pull the cheek and the upper lip downwards in order to precisely outline the working limit between the mobile and immobile mucous membrane (what determines the height of the obturator), and the position of the labial frenulum and other creases. The procedure and obturator making was simmilar to RB obturator made for the baby who had isolated cleft of secondary palate.

After the impression procedure having been completed, a working cast is made in the lab. At this development stage, the baby's alveolar ridges have not fully developed so they are called alveolar margins. The maxillary alveolar ridge is horseshoe-shaped and of a constant configuration. It develops in two parts: the exterior or labio-buccal and the interior or lingual part. The labio-buccal portion is differentiated first and grows more rapidly. That fact should be kept in mind when making obturators, in order to allow the growth of the alveolar ridge.

Accurate impression, regardless of its discontinuity, clearly shows all other anatomic details (surfaces) typical for this stage of the baby's development, and which can serve as orthodontic retainers, or on the contrary, can destabilize the stimulator (Figure 3).

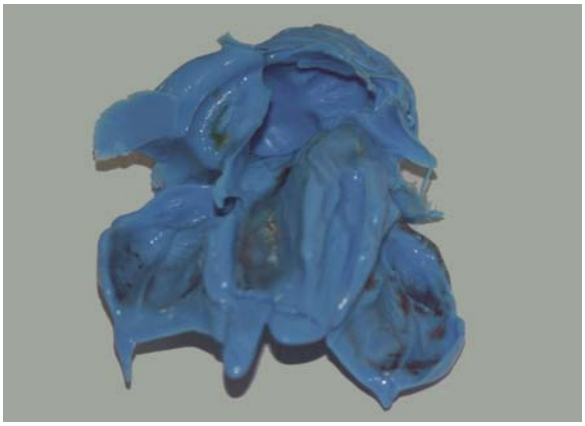

Fig. 3 – The second impression of the upper jaw and the soft palate.

At the next stage, the cleft space delineated on the working cast is filled with wax. That space results from the discontinuity in the fusion between the two palatal shelves. Using the working cast prepared in that manner, the RBJ obturator is made with polymerizing acrylates (Figure 4).

Fig. 4 – The first study model and the first RBJ obturator.

Its construction is almost completed by the polymerization of the acrylates, and the removal of sharp edges. In the end, the obturator is polished for a high shine to prevent a further damage to the mucosa in the baby's oral cavity. The obturator is then tried in the baby's mouth and checked for the accurate fit, while the mother is instructed how to use it when nursing her baby, and how to maintain the hygiene of the orburator (Figure 5).

Fig. 5 – Bottle-feeding of the baby.

Over the following treatment period, it is made every week for the first month due to the intensive growth of the baby, but later, it is done every three weeks. The role of the RBJ obturator is to separate the oral from the nasal cavity, what, thus, allow the creation of negative pressure, and enables baby's feeding.

At every two-week follow-up visit, the baby should be checked for the occurrence of lesions involving the soft tissue of the alveolar ridge (decubitus) or lesions on the frenulum that could be caused by the obturator. Then, the existing orburator is adjusted to the growth of the alveolar ridge by removing acrylate from the obturator's inner side (the side that lies on the alveolar ridge from the vestibular side).

In that way, the obturator prevents the cleft from further spreading as the baby grows, and, at the same time, allows positive growth of the alveolar ridges.

Once the baby is accustomed to the regular use of RBJ obturator, a slightly redesigned RBJ stimulator is made. Its purpose is to primarily close the separated segments of the alveolar ridge and palatum, and, thus, direct their further development, decrease the gap, and greatly facilitate the surgical reconstruction of the alveolar ridges and the palatum. This type of RBJ stimulator has an open orthodontic screw (bolt) that allows the active segments to move. By activating the screw, the segments move closer to each other (Figure 6).

Fig. 6 – The second study model and the second RBJ stimulator with a screw.

In order to avoid the discontinuity in the transverse development of the maxilla, acrylates are simultaneously removed from the side of the stimulator which is in contact with the vestibular side of the alveolar ridge. At the same time, this type of RBJ stimulator allows undisturbed feeding of a baby.

The fabrication procedure for this type is almost the same; it is made from the working cast but with a plate cut in two halves attached with an open screw. By its activation, the space between the halves of the plate decreases and the fragments get closer (Figures 4 and 6).

Two months after birth, a visible retroposition of the maxilla could be observed (Figure 7a) on the baby's face, as well as the improved position of the lateral segments when compared to the initial condition (Figure 7b). When necessary, this RBJ stimulator continues to be periodically fabricated.

Four months after the baby's birth, the position of the separated segments progressively improved, what also could be noticed on the baby's face (Figure 8).

Fig. 8 – The fourth RBJ stimulator, four months after birth.

By the time the baby was one year old, the separation distance between the fragments was so small that it could greatly facilitate the surgical treatment, and secure its success to a great extent (Figures 9 and 10).

Another great advantage of the RBJ stimulator is the absence of extraoral fixation. This appliance rests on the ridge of the upper jaw as a complete denture.

Fig. 7 – The third RBJ stimulator, two months after birth.
a) Visible retroposition of the maxilla; b) Improved position of the lateral segments.

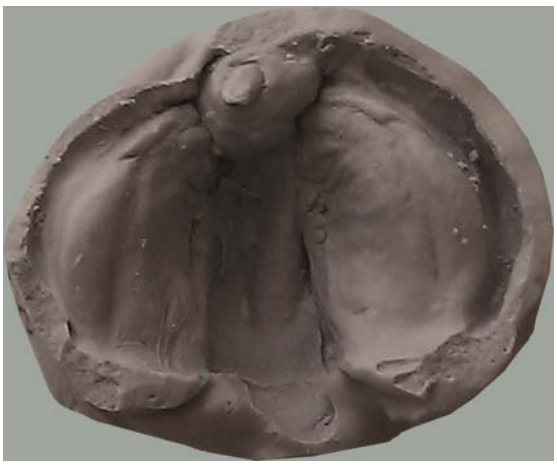

Fig. 9 – A plaster model of the 1-year-old baby's upper jaw.

Fig. 10 – The facial appearance of the 1-year-old baby with complete bilateral cleft lip and palate.

Babies are accustomed to its presence in the mouth, so that they always protest when the stimulator is removed from their mouths.

Discussion

In recent years, there have been numerous studies that questioned the validity of early application of orthodontic therapy in babies with cleft, primarily because they allow natural feeding. The most significant are the studies which have proved that the use of palatal obturators could shorten the feeding time, increase the milk intake quantity, and, as a result of all the mentioned above, allow a normal baby's growth and development. Different positions of a baby that could facilitate and ensure successful direct breastfeeding have also been studied.

The second, not less important reason for the application of early orthodontic therapy is early orthodontic treatment of the cleft. The presurgical infant orthopedic surgery as a neonatal therapy for the correction of cleft lip and palate was introduced in the mid-20th century. Some of the problems to which the traditional approach is applied include deformities of the nasal cartilage in both unilateral and bilateral cleft lip and palate cases, and deficient collumela in children with bilateral cleft. Early therapy for cleft lip, alveolus and palate achieves almost normal anatomical relations.

Descriptions of different intra- and extraoral appliances used for that purpose may be found in the literature.

Active intraoral appliances were described by Reisberg et al. [16].

Some authors directed the growth of the premaxilla in the downward and backward direction by application of force on the premaxilla [17–20].

Millard et al. [20] suggest that any application of force could have a restriction impact on the growth of the premaxilla.

Hotz et al. [21] thought that passive intraoral appliances should direct skeletal growth in the desired direction, or even stimulate the growth, what Weil [22] and Nolst et al. [23] confirmed in their studies.

Both active and passive appliances incorporaded to the lateral segments, provide a stabilizing effect on them. That was considered an advantage of those appliances.

Oosterkamp et al. [14] analyzed dimensions of the maxilliary alveolar arch in the patients with bilateral cleft lip and palate. Prior to the surgical closure of the cleft lip, children underwent early orthodontic therapy with intraoral retrusion plates. In 14 children with this anomaly (9 boys and 6 girls) born between 1999 and 2002, the effects of the application of those plates were assessed on plaster models cast from the maxillary impressions. The impressions were taken of the infants' upper jaws, and the passive feeding plates were fabricated. Three weeks later, the impression procedures were repeated, and new retrusion plates were made. These plates contained a microexpansion screw which was positioned in the way to allow movement into two directions: the retrusive movement of the premaxilla, and, if necessary, the realigning movement of the premaxilla relative to the vomer. This position was determined by clinical assessment of the casts. The front plate of the appliance over the premaxilla was connected through the screw with the second plate placed over the lateral segments.

The parents were instructed to turn the screw daily for a quarter of a cycle (0.175 mm/day), producing the retrusion of 2.5 to 3.7 mm in two to three weeks' time. Every three weeks, a new plate was fabricated, and the procedure was repeated until the time of surgical closure of cleft lip, i.e. when the baby reached 4 months of age.

The results indicated that during the therapy there was a significant decrease in the distance between the premaxilla and the lateral segments. That decrease correlated with the increase in the deviation of the premaxilla in relation to the vomer.

For each millimeter decrease in the distance between the premaxilla and the cleft lateral segments, an average increase in deviation of 4.0 degrees was found.

There was also a significant reduction in the width of the left and the right cleft, and a significant increase as well in the premaxilla width, while the transverse dimensions of the premaxilla did not significantly change in the area of lateral segments.

Active presurgical treatment with an oral retrusion plate proved to be very successful in orthodontic treatment of children with bilateral cleft lip and palate.

Abu-Rub et al. [24] studied the effects of an intraoral implant appliance on the reposition of dislocated segments of the upper jaw, as well as the magnitudes of palatine changes dependent on the age of babies born with bilateral cleft lip and palate. By using the 12 plaster study models, the condition of the babies (2–3 weeks after birth) prior to the introduction of the therapy, and their achieved condition at the time the retention (immediately before the cleft lip surgery) was assessed. The intercanine and the intertuberosity width, the palatal length and depth and the intercanine arch length were measured. The authors found an increase in the intercanine and intertuberosity width, and a reduction in the palate and the intercanine arch length. However, the authors did not found any link between these changes and the age of babies.

Spengler et al. [25] estimated the outcomes of presurgical alveolar molding in the therapy of patients with bilateral cleft lip and palate. Their study included the group of eight patients that were treated during the 2002–2004 period. The orthodontic therapy was initiated 34.9 days after birth, and lasted for the average period of 212.5 days. Following the impression procedure (performed under general anesthesia), the appliance was fabricated.

A package for the throat was used to prevent the unwanted entry of alginate materials into the oropharynx. The appliance consisted of acrylate palatal plates and two nasal stents. The stents were added to the appliance only after the intersegmental space had been reduced to less than 6 mm. Repositioning of the premaxilla between the lateral segments was carried by the application of outer force with the aid of sticking plaster and rubber bands, with which the appliance was attached to the child's face. In seven months' time, the columella was lengthened enough, the prolabium was wide enough and rotated inward to the maxillary arch.

The values obtained prior to the initiation of the therapy and after the treatment were compared on the basis of the cast measurements and statistical analysis. The results of the therapy recorded on the upper jaw molds showed that a statistically significant reduction of the premaxillary protrusion and the width of a bigger cleft were achieved. This improvement was comfirmed by the reposition of premaxilla and its alighment with the alveolar segments. However, the width of a smaller segment increased in a half of the studied children as the result of the maxillary repositioning, which was achieved through the width reduction of the bigger cleft.

The authors quantitatively showed that presurgical nasoalveolar molding had significant advantages in bilateral cleft treatment. The shape of the maxillary arch considerably improved after the alignment of the protruded premaxillary segment with the alveolar arches. As a result, the changes coupled with the orthodontic therapy helped reduce the surgical complexity.

Over the 12.5 year period, Garfinkle et al. [26] conducted a trial involving 77 infants born with non-syndromic bilateral cleft lip and palate. The children underwent an early orthodontic therapy with a nasal alveolar denture. Based on the used casts and the measurement results obtained with a digital caliper, the distances between five reference points were measured through the nasal structure. Those measurements were carried out at five different stages. No significant differences between healthy children and children with bilateral clefts were found.

Early presurgical orthodontic therapy has been used in more than 54% cleft palate centers in the world, including our previous case report on the application of palatal RB obturator in babies with isolated palate cleft. Regardless the fact that the results of the above-mentioned studies and this case report proved it to be useful and recommendable, there are, even today, doubts about its viability.

Conclusion

Based on the changes on the working casts monitored through the entire treatment period from the baby's birth up to one year of age, we could conclude that the application of the RBJ stimulator helped in progressive reduction of the cleft between the separated fragments and the premaxillary retrusion as well, in this severe case of complete bilateral cleft lip and palate.

One year after birth, a contact between the premaxilla and the lateral segments was established. That could not be achieved in any other way, but through the use of a stimulator. It further facilitated the surgical procedure to a great extent.

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