

How various surgical protocols of the unilateral cleft lip and palate influence the facial growth and possible orthodontic problems? Which is the best timing of lip, palate and alveolus repair? Literature review

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SUMMARY

Objectives. Cleft lip palate is congenital growth disease with unknown etiology, probably linked to both genetically and external causes. The aim of this work consists in presenting the effects of these diseases on cranio facial growth and the surgical protocols described in literature.

Materials and methods. The literature review articles conducted by Medline ranged from 1998 to 2011 have been selected. The key words of the research were “cleft lip palate”, “cleft lip palate facial growth”, “cleft lip palate surgery”. The inclusion criteria were articles that analyzed surgical protocols and the growth of unilateral lip and palate clefts, the timing repair of lip, palate and alveolus. We excluded case reports, studies without control group in the sample and the other types of publication as thesis or conference presentation. 60 articles had the selection criteria of the research.

Results. The cleft lip and palate is one of the most common birth defects that needs long rehabilitation between birth and adulthood. Several authors have presented surgical protocols and timing. The effects of these diseases on cranio facial growth and the importance of the early intervention have been described.

Conclusions. The review describes the main surgical protocols and treatment strategies of the unilateral lip and palate clefts. The review discusses how surgery effects the midfacial skeletal growth. Studies agree that the palate repair is the main cause of the maxilla growth disturbances. About the timing of palate repair in the unilateral clefts it can be concluded that most studies found no difference between one or two stages palate repair techniques for the midfacial growth. Also from the research, studies agree that delayed hard palate repair has more positive effects on maxillary growth than that of early hard palate repair. Nevertheless good results, delayed hard palate repair technique is abandoned by many hospital centres because of worse speech outcome. The best technique of palate repair is difficult to conclude, because the research results are part of a big controversy between the centers. From the studies about the alveolar repair it can be concluded that the primary bone grafting had more negative results on the skeletal growth. Gingivoperiosteoplasty and the secondary bone grafting had more positive results for intracranial relationship. The studies agree that the best timing of lip repair is during third-sixth month of life and that lip repair could have negative influence on the maxillary growth.

Key words: cleft lip palate facial growth, cleft lip palate surgery.

INTRODUCTION

Cleft lip and/or cleft palate is a congenital deformity that forms between fourth and ninth weeks of pregnancy. About 1 in 700 children are born with a cleft lip and/or cleft palate. Cleft lip forms because of the maxillary and medial nasal processes fusion failure. Cleft palate forms when two lateral palatine processes of hard palate are not completely fused. The etiology is multifactorial and the causes of cleft lip

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and palate are unknown, but are thought to be caused by the combination of genetic and environmental factors. Other causes could be some medicine somministration, exposure to virus during the pregnancy. The women who smoke or/and with diabetes have an increased risk to have a child with cleft lip and/or palate. Orofacial clefts can be categorized into three categories: cleft lip alone, cleft lip and palate (cleft extends from the upper lip to the hard or soft palate) and cleft palate alone. It can also be median, lateral and oblique. A cleft can occur on one side of the mouth and it is called unilateral cleft or on both sides – is called bilateral cleft. Complete cleft palate involves the entire primary and secondary palates. Incomplete cleft palate involves only secondary palate. There are various types: bifid uvula, submucosal cleft, soft palate cleft, soft and hard palate cleft. Two thirds of all clefts include cleft lip with or without cleft palate and one third of clefts are isolated cleft palates. In non-treated cleft lip and palate patients the midfacial growth is very similar when compared with non-cleft patients without apparent restriction of the growth. In the cleft patients the main cause of growth disturbances, especially the maxillary retrusion, is because of the chirurgical lip and palate repair. The study of H. A. Scheuer (1) concluded that in the cleft patients the reduction of the intermaxillary relationship (ANB) was because of midfacial growth impairment and it was not influenced by mandibular growth. The 4. 1° reduction of SNA between 8 and 16 years was found. The authors underlined that it is very important during the main orthodontic treatment period to decide if conservative orthodontic treatment can make dental compensation possible with functionally stable occlusion or it will be impossible and the orthodontic treatment should be delayed until orthognathic surgery. The study of Y. F. Liao (2) found a significant correlation between the cleft size and the growth of the maxilla – patients with the large cleft had a more retrusive maxilla. The study of M. Meazzini (3) analyzed the factors that could inhibit maxillary growth in cleft patients. The authors discovered that the most important factor that causes maxillary hipoplasia is congenitally missing lateral incisors. A cleft lip and palate can produce a variety of dental problems – number, size, shape, and position of the deciduous or permanent teeth. The teeth most affected are in the cleft area. The main orthodontic problem in unilateral cleft lip palate is maxillary asymmetry with the tendency of the cross bite formation. The cleft that involves the alveolar arch presents greater orthodontic problems and early orthodontics is essential. There is a discussion of the controversies of the cleft lip and palate

surgical techniques. The Euro cleft study showed that among the 201 European centers, 194 different treatment protocols existed for unilateral clefts (4). A team of medical professionals best treats cleft lip and palate. Usually the team consist of the surgeon, paediatrician, otolaryngologist, geneticist, psychologist, orthodontist, dentist, speech-language pathologist, audiologist. The treatment begins soon after the child's birth and continues until adulthood. The purpose of cleft treatment is aesthetic and functional rehabilitation. The surgical repair is important for facial growth preservation, normal speech formation and development of proper dentition. The less number of interventions, the less the scaring results and, hence, growth retardation (5). The purpose of this study was to evaluate the main surgical protocols and the best timing of the unilateral cleft lip and palate. It was concentrated in particular on the cleft palate, alveolus and lip repair and how the surgery affects the facial growth and development.

MATERIALS AND METHODS

The articles for the systematic review were taken from the electronic database Medline (www.ncbi.nlm.nih.gov/pubmed). The key words of the research were “cleft lip palate”, “cleft lip palate facial growth”, “cleft lip palate surgery”. The articles were selected only which were written in English from 1998 to 2011 years. The inclusion criteria were articles that analyzed surgical protocols of unilateral lip and palate clefts, the surgery effects on the facial growth and facial skeleton development. The articles about orthodontic problems in cleft patients that used for studies the cephalometry and the study models were selected and included in the review. Publications about the timing repair of lip, palate and alveolus were also researched and included in the review. We excluded case reports, studies without control group in the sample and the other types of publication as thesis or conference presentation. 60 articles had the selection criteria and were analyzed out of a research of 382 articles.

RESULTS

Palate repair

There are different opinions about the ideal technique of palate repair and timing. Nevertheless there are many surgical techniques and modifications, all protocols focus on speech development, midfacial growth and velopharyngeal function. To achieve these goals, repair of the cleft palate must include closure in three layers, including a nasal layer, muscle layer and

oral mucosal layer. Compromising closure of any of these layers increases the incidence of postoperative complications (6). There are two main techniques of the palatal clefts closure depending on timing (7). The first technique is two-stage protocol that consists of two separate operations. The first serves to close soft palate and the second serves to close the hard palate. The hard palate repair can be delayed until mixed dentition. The main advantage of this technique is to avoid disturbances on facial growth. The main disadvantage is impairment on the speech development. The second technique is one stage repair, when the soft and the hard palate are closed at the same time. The negative aspect of this protocol could cause disturbances on the facial growth.

Of all 60 publications 26 were about surgical protocols of palate repair and timing. Of these 26 articles, 10 articles were about surgical protocols of palate repair by stage one or stage two and the differences on midfacial growth. The review of D. S. Precious (8) suggests soft palate to close around at 6 month and hard palate around 12 month of life and does not recommend techniques that involve medial displacement of the palatal fibro mucosal flaps. Three studies of ten showed that two-stage protocol have more advantages on midfacial growth (9-11). T. Yamanishi from his study of early two stages and early one stage palate repair with two different protocols found that two stages protocol is advantageous for maxillary growth and speech development. He suggests that critical period of hard palate closure for adequate speech development is before 3 years of age. Two studies (12, 13) concluded that one stage protocol is better for the maxillary growth respect two stages protocol. The study of W. Pradel (12) found that one stage early repair at the age of 9-12 months has a more positive influence on speech development and early maxillary growth than that of early two stage palate repair. Five studies found no significant differences between two protocols of palatal repair (14-18). The study of M. Corbo (14) used one stage protocol and two stages protocol of palate repair for comparison of the craniofacial development. In one stage operation the lip and palate were closed at 3 months of age and in two stages – soft palate was closed at 3 months, lip and hard palate at 6 months. The study discovered no differences on craniofacial dimensions between two groups. The study of W. Zemann (15) found very similar sagittal growth of midfacial when comparing two surgical protocols at 6 years old children. The two techniques were: first - one stage palate closure at 12 months of age according to Veau technique and second – two stage palate closure with soft palate closing at 12 months

and hard palate at 2. 5 years of age. At the second study of W. Zemann (16) other evaluations were performed at the age of 10 years. It only confirmed that there were no significant differences in sagittal growth between two techniques. Nine studies out of 26 were about delayed hard palate closure and the influence on facial growth (10, 11, 13, 17-23). Supporters of this technique think that delayed hard palate repair has positive effects on maxillary growth; usually it is delayed until the mixed dentition. The studies of Yu-Fang Liao (10, 19) found that late hard palate repair has a smaller adverse effect than that of an early hard palate repair on the maxillary growth. His publication (10) studied two groups with different surgical protocols. In the first group palate was closed by one stage around 1 year of age and the second group had two-stage palate repair, while soft palate was closed around one year of age and hard palate at six years of age. This study demonstrated that the delayed hard palate repair is more advantageous on the growth of the maxilla and on the anteroposterior jaw relationship. It was also confirmed that palate repair has no effect on the downward growth of the basal maxilla and the growth of the mandible. The study of H. Friedy (21) concluded that two slightly different surgical protocols of Goteberg and Riga with delayed hard palate repair gave the good midfacial development. The studies of P. J. P. M. Nollet (20) and J. Dask (17) found that the delay in hard palate closure did not result in improved maxillary growth. On the contrary of the other studies, A. Gaggl's study (13) concluded that a more severe impairment of the maxilla growth in the sagittal and frontal plane was observed after two stage with delayed hard palate repair than with an early one stage technique. Even though there are good results on maxillary growth of delayed hard palate repair, this technique is highly being abandoned. The main reason is speech defects with velopharyngeal insufficiency. There are many surgical techniques and many variations of cleft palate repair. The most famous techniques are:

- *Von Langenbeck palatoplasty* – is probably the oldest. The cleft is closed by mobilizing bipediced mucoperiosteal flaps medially. This technique is ideal for isolated cleft palate repair. It is used today with the modification and in combination with other techniques. The three studies of all the palate repair publications were about Von Langenbeck technique (24-26). The study of M. Kulewicz (24) compared three different surgical protocols. At 7 months of age patients underwent one stage surgery that consisted of lip, soft and hard palate repair.

Hard palate repair was different – in the first surgery hard palate was closed according to the bilateral Von Langenbeck's technique, in the second – according to the unilateral Von Langenbeck technique and the third – using a single-layered caudal-pedicled vomer flap, which limits the hard palate mucoperiosteal flap elevation. This study showed that the third surgery group had the most favourable craniofacial morphology. In the study of R. W. Pigott (25) there was also the comparison of three different surgical techniques of hard palate with identical management of lip, nose, alveolus and soft palate. At 6 months, patients had three-layer repair of soft palate with different residual hard palate closure – Cuthbert three-flap modification of the Veau technique, Von Langenbeck technique and the medial Langenbeck procedure. The results showed significant differences among three groups with more favourable anteroposterior maxillary growth of the third group.

- *Veau-Wardill-Kilner or VY pushback palatoplasty* – it can be used to increase the anteroposterior length of the palate that improves velopharyngeal competence, because mucoperiosteal flaps are retroposed and palate is lengthened. The disadvantages are the denuding palatal bone anteriorly that can negatively influence midfacial growth. The hospital centers are abandoning this technique. From the palate repair publications, the five studies examined the Veau technique (13, 27-30). The study of S. Choudhary (27) concluded that satisfactory long-term midfacial growth could be obtained with Veau cleft palate repair. The final Goslon results showed that 72% of the patients had a good outcome. The publication of F. Farzaneh (29) studied the effects on the morphology of the skeleton and dental occlusion of two different surgical protocols – Von Langenbeck and Veau-Wardill. The study found the similar facial morphology outcome of two surgical techniques. The study of M. J. Gaukroger (28) compared two methods: Mount Vernon hospital method and Oslo hospital method. The hospital of Mount Vernon used Veau-Wardill technique and Oslo hospital used Von Langenbeck technique. M. J. Gaukroger found that patients of Mount Vernon presented a more flat facial profile and had more reduced maxillary prominence.

Bardach's two-flap technique – is used to repair complete clefts. The incisions are made along the cleft margin and the alveolar ridge margin with elevation of two mucoperiosteal flaps. Both are done by intravelar veloplasty.

- *Furlow double opposing Z-palatoplasty* – involves two reversed Z-plasties based on the cleft midline continued in soft palate. The nasal and oral flaps are mobilized posteriorly. The advantage is that soft palate can be lengthened. The study of T. Yamanishi (9) compared two surgical protocols – stage two double opposing Z-palatoplasty and stage one Veau-Wardill palatoplasty. The study found more advantageous maxillary growth and speech development of stage two double opposing Z-palatoplasty.
- *Vomer flaps* – there are many variations of the vomer flap. They are used in unilateral and bilateral clefts for nasal lining or oral mucosa resurfacing. The most used for bilateral clefts is the four-flap palatoplasty.
- *Intravelar veloplasty* – is one of the techniques to repair the soft palate. By repositioning the elevator muscle you achieve the velopharyngeal competence. This technique is widely used today. The older standard for velopharyngeal insufficiency was the side-to-side technique by Veau. The study of B. Richards (31) compared two surgical protocols. The first surgical protocol of the Oslo hospital, which repaired lip and hard palate at 3 months and the soft palate at 12 months of age. The second surgical protocol of the Paris hospital, which closed the soft palate at 3 months and at 6 months of age closed the lip and hard palate. It was concluded that there was no significant difference in facial growth between two types of repair.

Cleft Alveolus repair

Cleft alveolus is a maxillary alveolar ridge bony defect that is presented in about 75 % of cleft lip palate patients. There is still controversy about the surgical techniques and the best timing of the cleft alveolus. A total 13 publications were about cleft alveolus and treatment solutions. There are three surgical solutions to repair cleft alveolus: gingivoperiosteoplasty, primary bone grafting and secondary bone grafting. Gingivoperiosteoplasty- is also called boneless bone grafting. It is common today to use it with presurgical orthopaedics because it helps to decrease cleft width. The most common timing of gingivoperiosteoplasty is at the same surgical operation when the lip repair

is being performed. The advantages are that it reduces the need for the secondary bone grafting because it allows the growth of new bone and enables the eruption of the permanent teeth. There are four studies that performed gingivoperiosteoplasty simultaneously with palatoplasty (32-35). All of them present good facial growth results. In the study of W. D. Losquadro (32) with the protocol of New York, gingivoperiosteoplasty was being performed with palatoplasty at 12 months of age. The results showed no significant growth disturbances and good bone growth that reduced the need of secondary bone grafting. The study of J. N. Mcheik (34) presented early repair of cleft when rinocheiloplasty by Millard technique was performed at 1-4 weeks and at 6 months were performed palatoplasty with gingivoperiosteoplasty. The study showed the good facial growth and aesthetic results. Primary bone grafting – is performed before 24 months of age and the main advantage is that it stabilizes maxillary arch with prevention of the arch collapse. It also creates more uniform growth of the maxilla with improvement of articulation. The disadvantage is that it can provoke attenuation of the maxillary growth especially in the vertical dimension. The standard bone graft used is rib autograft. It is important not to perform the surgery in the area of vomeropremaxillary suture, because it can cause the impairment of maxillary and midfacial growth (36). Five studies were discussed about primary bone grafting (36-40). Three publications of them found more negative results of primary bone grafting compared with the groups, which underwent other surgical protocols. The study of G. T. Sameshima (37) compared two groups, first operated at 8 months by cheiloplasty with gingivoperiosteoplasty and the second at 7 months by cheiloplasty with primary bone grafting. Two groups underwent delayed palatoplasty at about 4-5 years of age. The results showed that the first group had a more horizontal growth of the mandible with less large symphysis. The study of K. Maresova (40) concluded that the development of the intracranial relations is more positive in patients who underwent gingivoperiosteoplasty compared to the patients who underwent bone grafting. The mandibular growth leads to the posterior rotation in the patients with bone graft. One of the studies of R. Hathaway (36) compared the craniofacial growth of two groups of patients – the first group underwent primary bone grafting and the second without primary bone grafting. It was discovered that the patients of the first group presented a more frequently maxillary retrusion and mandibular opening. The other study of R. Hathaway (38) compared arch dimensions of two groups - one with primary bone grafting and the

other without primary bone grafting. He found no significant difference for any arch dimension-length and width between the two groups. Of all cleft alveolus publications, one study that was performed by S. W. Rosenstein (39) was speaking about the advantages of results of primary bone grafting. The protocol includes early maxillary orthopaedics with primary bone grafting. Before lip repair, maxillary prosthesis is placed and later at 4 to 8 months a rib graft is placed across the alveolus. The study found a potential good maxillary growth and no significant difference compared to the group that did not undergo the primary bone grafting.

Secondary bone grafting – usually is performed before permanent canine eruption and involves the grafting of autogenous cancellous bone from iliac crest. The advantages are that it minimizes the growth disturbances of the upper arch, and gives maxillary arch integrity with periodontal support for the teeth proximal to the cleft. Secondary bone grafting is now widely used and is considered a standard procedure for alveolar repair. Four studies examined the secondary bone grafting (3, 8, 35, 41). All of them were speaking about the comparison of two surgical protocols – Milan and Oslo. Milan's protocol include lip, nose and soft palate repair at 4-6 months of age and hard palate repair with gingivoperiosteoplasty at 18-36 months of age. Oslo's protocol includes lip and anterior palate closure by vomer flap at 3 months and soft palate closure at 18 months of age. The secondary bone grafting is performed at 8-11 years, before canine eruption. The results at 5 years of age showed no significant differences cephalometrically between two groups and at 10 years of age there was a better outcome of the maxillary growth of the group with secondary bone grafting with less need for orthognathic surgery. The review for dentist of D. S. Precious (8) recommends the secondary bone grafting and concluded that the best timing of cleft alveolar bone grafting is early mixed dentition stage for the periodontal health improvement of the permanent erupting dentition.

Cleft lip repair

Usually the nose correction is performed simultaneously with lip repair. A primary lip repair is an important factor for symmetrical outcome of the nose. The reconstruction of perioral and perinasal muscles has been considered especially crucial to allow undisturbed development after surgery (21). Surgery of cheiloplasty is usually performed during the third – sixth month of life and there is not the big controversy of optimal timing as for the palate repair between hospitals. There are several techniques of

cheiloplasty. The most frequently used are: Rotation advancement by Millard, Triangular techniques (by Tennison with modifications), wave line closure by Pfeifer, lip closure by Delaire. The main difference of the lip repair techniques – Millard, Tennison, Pfeifer are the different line incisions. The Millard rotation advancement is probably the most widely used. The advantages are that this technique repairs a normal-looking cupid's bow, where the minimal amount of tissue is removed. There are many adaptations and variations of this technique. The cheiloplasty by Delaire takes more attention to reconstruct different muscle groups and is called functional repair. There is a controversy in the literature if surgical repair of the lip could restrain the maxillary growth. Pressure from a tense upper lip might cause maxillary anterior teeth to become retroclined, usually resulting in an anterior cross bite situation (21). Although there is an agreement that palate repair causes the maxillary growth disturbances. Six studies examined the techniques of lip repair and the influence on the maxilla growth (26, 42-46). The study of Yang Li (42) compared two groups: the first group had at 9 months of age only labioplasty and the second group had both lip repair at 9 months and palate repair at 36 months of age. The results of this study showed that maxillary retrusion in both groups were identical. The study concluded that lip repair is the most important factor that restrains maxillary growth. The study of Yu-Fang Liao (26) compared two groups: the first group with the lip and palate repair and the second group only with the lip repair. He concluded that palate repair restrains the development of the maxilla because he found that the first group had a smaller maxillary length and an anteroposterior jaw relationship compared to the second group. The study of G. Schultes (44) compared midfacial growth of two groups: the first group of patients with clefts of palate and the second group of patients with unilateral clefts of lip, palate and alveolus. The study concluded that the group of isolated clefts palate showed better cephalometric results. The group of unilateral clefts showed more retrognathic face with transverse asymmetries. The study of C. S. Huang (43) examined how lip repair affects the maxillary dental arch development and found that cheiloplasty by Millard could give continuous pressure to the anterior maxillary arch. The study of R. Rullo (45) examined the effect on midfacial growth of Delaire's cheilorhinoplasty. He underlined that the closure of the cleft lip could be responsible for maxillary retrusion and it was important to rebuild carefully the perilabial muscles to have the positive maxillary growth. The review for dentist of D. S. Precious (8) suggested the end of sixth month for

the optimum timing of cheiloplasty, because early reconstruction of the lip inhibits the development of the premaxilla and reduces forward growth.

Discussion and Conclusion

The cleft lip and palate is one of the most common birth defects that needs long rehabilitation between birth and adulthood. The review speaks about the main surgical protocols and treatment strategies of the unilateral lip and palate clefts. It is discussed how surgery affects the midfacial skeletal growth (46-60). Studies agree that the palate repair is the main cause of the maxilla growth disturbances that inhibits basal maxilla to displace forward and to develop anteroposteriorly. The cleft palate closure inhibits normal sutural activity that tends to create a class 3 deformity. It can be concluded about the timing of palate repair in the unilateral clefts that most studies (five out of ten studies that examine surgery protocols of palate repair by one or two stage) found no difference between one or two stage palate repair techniques for the midfacial growth. From the research, seven out of nine studies that examined delayed hard palate closure agree that, delayed hard palate repair has more positive effects on maxillary growth with smaller adverse effect than, that of early hard palate repair. Nevertheless good results delayed hard palate repair technique is abandoned by many hospital centres because of worse speech outcome. Despite wide variation in the timing of hard palate repair in current use, according to the survey of the euro cleft project, more than 90% of the 201 registered centres complete hard palate closure before 3 years (4). The best technique of palate repair is difficult to conclude, because the research results are mixed by big controversies between the centres. From the 13 studies about the alveolar repair, five spoke about the primary bone grafting. The three publications found that the primary bone grafting had more negative results on the skeletal growth. Four studies were about gingivoperiosteoplasty. All of them reported good facial growth results. Four studies examined the secondary bone grafting and all of them had better outcome of the maxillary growth with more positive results for intracranial relationship respect the gingivoperiosteoplasty. We can conclude that patients with secondary bone grafting in general have more positive intracranial relations. We advocate according to this study results the gingivoperiosteoplasty and secondary bone grafting for the alveolar cleft reconstruction to achieve function improvement and aesthetics. The six studies that analyzed lip repair agree that the best timing of lip repair is during third-sixth month of life and that lip repair could have the negative influence on the maxillary growth. In our

point of view according to this study we conclude that the lip reconstruction can reduce the growth of the entire dentoalveolar segment and underdevelopment of the premaxilla with the difficulty to correct it later. We consider that to achieve the normal maxillary growth, the orthodontic treatment is one of the most important factors as surgery, where the main goal is the attentive reconstruction of the palate, lip

and alveolus, which ensure a more regular development of facial skeleton. This literature review can be useful to prepare dentists to recognize the functional and anatomic problems that create surgery of the unilateral clefts, to give more knowledge of the main surgical protocols and to explain the importance of the early treatment because of the existing dentoskeletal imbalance and facial asymmetry.

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