Pre-surgical orthopedics success in patients with cleft lip and cleft palate - Scoping review.

Cristhian Camilo Riveros Carvajal,1 Karla Xilena Calderon Guzman,1 Kimberly Yulieth Hurtado Cristancho,1 Cristhian Ariel Cisneros Hidalgo,2 Claudia Liliana Cabrera Arango.3

Abstract: Kerr McNeil, Scottish prosthetic introduced the Neonatal maxillary orthopedics two centuries ago setting the concept of using orthopedic parts to. Three different types of pre-surgical orthopedics have been described. These are: active, semi-active and passive. Objective: Perform a research in indexed data regarding the success of pre-surgical orthopedics treatment in patients with a condition of cleft lip and cleft palate. Methodology: A computer based research was performed at electronic data bases such as: PUBMED, SCIENCEDIRECT, PROQUEST, COCHRANE, OVIDSP SCOPUS, and MEDLINE. The Booleans researches used in the multiples data bases were OR, AND, NOT. Findings: Thirteen papers met the eligibility standards and were included in this review. 4 (four) of them are descriptive, longitudinal and prospective observational studies. 5 (five) are clinic reports, 1 (one) is a retrospective study, 1 (one) is a Cochrane review, and 2 (two) are clinic cases and check ups. The orthopedics treatment implementation depended on the kind of extension cases and severity of the cleft palate. Conclusion: the pre-surgical orthopedics is successful if it is implemented at birth up to 3 months of age. It assures satisfactory results decreasing considerably cleft palate, nasal malformation, and cleft lip before performing Cheilorrhaphy and Palatorrhaphy procedures.

Key words: Cleft lip, cleft palate, orthopedic surgery, palatal obturators, nose deformities.
Introduction

Congenital malformations, like cleft palate and cleft lips, are developmental disorders. They consist on the lack of fusion between the two segments of the lip in a single structure plus a defect in the development of the palate. They are characterized by the absence of complete fusion of the two palatal crests, resulting in a communication with the nasal cavity affecting the natural functionality of the stomatognathic system producing several side-effects such as physical and mental disorders in children and adults lives who suffer from this condition.\textsuperscript{1,2}

The etiology of oral fissures includes three facts; these are: environmental, genetic and ethnic elements. Among those belonging to environmental agents we find smoking, severe alcohol consumption, drug addiction, some medicine use, chemical and radioactive exposure, and infections.\textsuperscript{3}

The average cleft lip and cleft palate prevalence worldwide is 1 per 1,700 live births, being Japan the country with the highest rate 20.0 per each 10,000 live births; followed by United States of America with 10.2 per each 10,000 live births.\textsuperscript{4} Meanwhile, the Colombian rate is 1 per each 500 or 1 per each 1000 according to the geographic zone.\textsuperscript{2} Many studies conducted in the last few years in Colombia have shown that congenital malformations are related to severe public health issues.\textsuperscript{5}

The etiology of oral fissures includes three facts; these are: environmental, genetic and ethnic elements. Among those belonging to environmental agents we find smoking, severe alcohol consumption, drug addiction, some medicine use, chemical and radioactive exposure, and infections.\textsuperscript{3}

The average cleft lip and cleft palate prevalence worldwide is 1 per 1,700 live births, being Japan the country with the highest rate 20.0 per each 10,000 live births; followed by United States of America with 10.2 per each 10,000 live births.\textsuperscript{4} Meanwhile, the Colombian rate is 1 per each 500 or 1 per each 1000 according to the geographic zone.\textsuperscript{2} Many studies conducted in the last few years in Colombia have shown that congenital malformations are related to severe public health issues.\textsuperscript{5}
alterations in number, size and dental shape, dental enamel hypoplasia, tooth decay, endodontics related to the cleft, ectopic eruption, delayed dental eruption, and periodontal defects such as: gingivitis, periodontitis, cicatricial retractions, and bifid uvula. 6

There are multiple categories of cleft lip and palate. The Veau and Kernahan classifications reported by Monasterys L stand out, classifying them as follows: 1: Defects of the soft palate only. 2. Defect wrapping the soft and hard palate reaching the incisive foramen, 3: Complete unilateral cleft going from the soft palate to the alveolus reaching the lip, 4: Complete bilateral cleft, pre-maxilla is suspended from the nasal septum. Davies and Ritchie were the first to describe the submucosal cleft in 1922. The Kernahan and Stark classification includes: 1. Primary cleft palate only. 2. Secondary palate only slit. 3. Primary and secondary cleft palate. Kernahan designed a schema where we can draw the cleft in order to determine how severe the condition is as well as its classification reflecting the affected structure. 7

The diagnosis of cleft lip and cleft palate is performed through clinical exploration of the baby at birth. However, these malformations are also detected by transvaginal ultrasound, fetoscopy, and amniocentesis. There are also different types of treatments that require a more multidisciplinary handling to improve or decrease the cleft conditions divided into two big areas: surgical and non-surgical. 8 Surgical option embraces maxillofacial a plastic procedures and anesthesia. Among the nonsurgical specialties involved in the care of the cleft patient are: stomatology, pediatric dentistry, otorhinolaryngology, phon audiology, pediatrics, psychology, medicine, genetics, orthodontics, social work, nutrition. 9

The non-surgical treatment, includes presurgical orthopedics which is a procedure based on maxillary functional orthopedic plates that will favor plates that foster stimulation and bone remodeling of nasal and alveolar segments, and cleft palate. 13 This decreases the fissure wideness during the first months of life. It allows developing a more natural-like anatomy prior to surgery. The ultimate aim of functional orthopedic plates and nasal remodeler (NAM) consists of promoting and redirecting the growth of oral and nasal structures. 14

Three types of pre-surgical plates have been described: active, semi-active, and passive. 10 Functional orthopedic plates stimulate the transverse and anteroposterior growth of the palate, and allow anchoring other attachments to improve the conformation of the affected nasal structures and which produces a separation between the nasal cavity and the oral cavity, that in patients with complete clefts of lip and palate can affect the upper lip, alveolar rim, soft and hard palate, premaxila, filtrum, columella, floor and the alar base of the nose and it causes difficulty swallowing and feeding, regurgitation of food through the nose, middle ear infections with hearing loss and hypernasal voice.

The Nasoalveolar molder is also frequently used. It consists in a device that helps to shape and correct the abnormal position of the nasal cartilage and the alar rim of the affected side giving symmetry to the
nose. This nasal conformer can be joined to the functional orthopedic plate or can be designed separately. In the newborn this system of traction is based on the natural condition of elasticity and plasticity of its anatomical structures. Due to this condition, the nasal ala can be modeled before surgery. To diminish the fissure and improve nasal symmetry, the nasal conformer is attached to the molder; this represents an alternative that contributes to minimize the evidence of this congenital defect. The procedure makes it possible to align and approximate the alveolar and labial segments. It is the union of the palatal obturator and the nasal molder in one. Based on what was said before, this type of review helps professionals to broaden their knowledge about the subject, thus focusing on a multidisciplinary treatment since all the different classifications can be identified as well as their clinical manifestations on children suffering from cleft lip and cleft palate.

This offers an alternative treatment with pre-surgical orthopedics in this type of malformations. Children with this condition are the main beneficiaries by enhancing their quality of life integrating them into society, improving their self-esteem, and providing parents with essential tools for their nutrition and care giving the family the important role it deserves in the process. Therefore, the aim of this review was to conduct a search in indexed databases on the success of pre-surgical orthopedics in patients with cleft lip and cleft palate.

Methodology

Inclusion criteria includes reports and case series, literature reviews and original studies with no language barriers and the date were the successful presurgical orthopedic treatment took place on patients with cleft lip and palate.

We considered as exclusion criteria those studies that report in any oral surgery in patients target of study (Cheiloplasty - Palatorraphy) before orthopedics, and articles that do not cite to the time of use.

Research: A computerized search was performed in the following electronic databases: PUBMED, SCIENCE DIRECT, PROQUEST, COCHRANE, OVIDSP SCOPUS, and MEDLINE. The search was conducted during the months of August and September 2018.

The Boolean search engines used in the different databases were OR, AND, NOT. The following search strategy was performed in each of the databases with the following keywords: Cleft lip, Palate AND, orthopedic treatment, OR, treatment pre-surgical OR, Nasoalveolar molding OR, blanking and stimulating plate OR, bilateral cleft lip reconstruction.

Article choice: It was first done by taking into account the title, and summary of the papers. Those that did not show any match were excluded immediately.

Data extraction: It was carried out by the researchers independently, with the collaboration of the consultants in charge of the research. The following data were extracted: year of publication, name of the author, title of the article, treatment period, objective of the article, results, and conclusions. As shown in the following chart on unit of analysis (table 1).
### Table 1. Unit of analysis

<table>
<thead>
<tr>
<th>Title</th>
<th>Author(s)</th>
<th>Date of publication</th>
<th>Objectives</th>
<th>Type of treatment</th>
<th>Treatment period</th>
<th>Conclusions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Presurgical nasoalveolar moulding: A boon in the management of cleft lip and palate</td>
<td>Chaudhary, D. C et al.</td>
<td>Sep 19/2015</td>
<td>To evaluate the success of the nasoalveolar molder.</td>
<td>Nasoalveolar molder and palatal obturator</td>
<td>First week of birth at 2.5 months</td>
<td>The nasoalveolar molder is highly recommended for reducing the gap and nasal asymmetry.</td>
</tr>
<tr>
<td>Comparative Study of Nasoalveolar Molding Methods: Nasal Elevator Plus DynaCleftt Versus NAM-Grayson in Patients With Complete Unilateral Cleft Lip and Palate</td>
<td>Monasterio, L., et al.</td>
<td>June/2012</td>
<td>To compare the dynaCleftt nasoalveolar molder with the molder Grayson.</td>
<td>Nasoalveolar moulder</td>
<td>Day 14 of birth at 3 months</td>
<td>Both molders are beneficial for patients with cleft lip and palate, the authors of the article highlight the use of the DynaCleftt one for the different advantages it posses.</td>
</tr>
<tr>
<td>Efficacy of the nasal molding in patients with unilateral cleft lip and palate in newborn to 6-month-old patients</td>
<td>Restrepo, N. L et al.</td>
<td>December /2016</td>
<td>To evaluate and describe the clinical changes of the nasal ala with depression and Asymmetry conditions in patients with cleft lip and palate.</td>
<td>Nasoalveolar molder and palatal obturator</td>
<td>Patients between 0 and 6 months with a treatment between 45 and 192 days</td>
<td>Nasoalveolar molder and palatal obturator help improve nasal depression and cleft palate by up to 50%.</td>
</tr>
<tr>
<td>Presurgical orthopedics in patients with unilateral cleft lip and palate: clinical case reports</td>
<td>Rivas, D. D. et al.</td>
<td>June/2017</td>
<td>To evaluate the efficiency and time of use of stimulatory palatal obturator</td>
<td>palatal obturator</td>
<td>Patient 0 to 8 months old for 4 months of treatment</td>
<td>Pre-surgical orthopedics with the stimulating palatal obturator is an acceptable and recommended therapy for patients with cleft lip and full unilateral palate, the time of use of the orthopedics was 2 months obtaining good results.</td>
</tr>
<tr>
<td>Pre-surgical orthopedics in newborn patients with cleft lip and palate</td>
<td>Restrepo, N. L et al.</td>
<td>March/ 2016</td>
<td>To describes 2 cases of newborn patients who went through pre-surgical orthopedics.</td>
<td>Nasoalveolar molder and palatal obturator</td>
<td>Between 8 and 16 weeks of treatment</td>
<td>It is a successful alternative in the closure of the cleft lip and palate, demonstrating in two clinical cases the positive effects, and stressing the importance and accompaniment of parents.</td>
</tr>
<tr>
<td>Influence of nasoalveolar molding on skeletal development in patients with unilateral cleft lip and palate at 5 years of age</td>
<td>Akarsu-Guven, B. et al.</td>
<td>April/ 2018</td>
<td>To assess the influence of pre-surgical nasoalveolar molding on skeletal development in patients with unilateral cleft lip and palate operated at 5 years of age.</td>
<td>Nasoalveolar molder</td>
<td>3 to 4 months of treatment</td>
<td>The nasoalveolar molder did not lead to any significant difference in skeletal development in patients with cleft lip and cleft palate</td>
</tr>
<tr>
<td>Title</td>
<td>Author(s)</td>
<td>Date of publication</td>
<td>Objectives</td>
<td>Type of treatment</td>
<td>Treatment period</td>
<td>Conclusions</td>
</tr>
<tr>
<td>----------------------------------------------------------------------</td>
<td>------------------------</td>
<td>---------------------</td>
<td>-----------------------------------------------------------------------------</td>
<td>----------------------------------------------------------------------------------</td>
<td>------------------</td>
<td>-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------</td>
</tr>
<tr>
<td>Presurgical Nasoalveolar Molding of Bilateral Cleft Lip and Palate Infants: An Orthodontist's Point of View</td>
<td>Altuğ, AT et al.</td>
<td>2017</td>
<td>To conduct a general review of pre-surgical infant orthopedics and its contribution to subsequent dental practice.</td>
<td>Nasoalveolar molder and bilateral palatal obturator</td>
<td>Patients 1 week old with a treatment for 3 months</td>
<td>The pre-surgical nasoalveolar molder and the gingivoperiosteoplasty have demonstrated pleasant and stable nasolabial aesthetic advantages with fewer scars, intact maxillary dental arch without oronasal fistulas and reduction in the number of soft tissue revisions and alveolar graft surgeries.</td>
</tr>
<tr>
<td>A Comparative Evaluation of Efficacy and Efficiency of Grayson’s Presurgical Nasoalveolar Molding Technique in Patients with Complete Unilateral Cleft Lip and Palate with Those Treated with Figueroa’s Modified Technique</td>
<td>Singh, A., et al.</td>
<td>2018</td>
<td>To compare the effectiveness and efficiency of Grayson technique with the Figueroa modified pre-surgical nasoalveolar technique in a completely unilateral cleft lip and palate in babies.</td>
<td>Palatal obturator</td>
<td>Neonates 10-15 days old with a 6-month treatment</td>
<td>There is a significant reduction in the horizontal symmetry (width of the nostril and basal width of the nasal fossa) and a significant increase in the vertical symmetry (height of the nostril, height of the dome of the nasal fossa and columnar height) There was a significant reduction of the alveolar gap and there was a significant increase in the width of the arch.</td>
</tr>
<tr>
<td>Presurgical nasoalveolar molding in unilateral cleft lip and palate</td>
<td>Attigupp, P. R. et al.</td>
<td>2015</td>
<td>To determine the naso-alveolar molder success in patients with cleft lip and palate.</td>
<td>Nasoalveolar molder and palatal obturator</td>
<td>2-month-old patient with 2-year follow-up treatment</td>
<td>Pre-surgical nasoalveolar molding PNAM easy and passive method to join the alveolus and the lips together by redirecting the natural growth.</td>
</tr>
<tr>
<td>Presurgical nasoalveolar molding: A boon to facilitate the surgical repair in infants with cleft lip and palate</td>
<td>Prabhakar Ramasetty, Attiguppe, et al.</td>
<td>2016</td>
<td>To evaluate the success of the palatal obturator with nasoalveolar molders.</td>
<td>Palatal obturator with unilateral and bilateral Nasoalveolar molder.</td>
<td>3-month-old patients with a 3-month treatment</td>
<td>Long-term studies have shown that changes in the nasal shape is stable with less scar tissue and better labial and nasal shape. This improvement reduces the number of surgical revisions for excessive scar tissue, oronasal fistulas, and nasal and labial deformities.</td>
</tr>
</tbody>
</table>

Table 1. Unit of analysis (cont.)
### Table 1. Unit of analysis. (cont.)

<table>
<thead>
<tr>
<th>Title</th>
<th>Author(s)</th>
<th>Date of publication</th>
<th>Objectives</th>
<th>Type of treatment</th>
<th>Treatment period</th>
<th>Conclusions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Presurgical Nasal Molding With a Nasal Spring in Patients with Mild-to-Moderate Nasal Deformity with Incomplete Unilateral Cleft Lip with or Without Cleft Palate</td>
<td>Peanchitlertkajorn, S. et al.</td>
<td>2018</td>
<td>To determine the benefit of using the nasoalveolar molder</td>
<td>Nasoalveolar molder</td>
<td>Neonatal patient with a treatment for 11 weeks</td>
<td>It proved to be effective, achieving good nasal symmetry and lengthening the nasal columella.</td>
</tr>
<tr>
<td>Presurgical Orthopaedic Nasoalveolar Molding in Cleft Lip and Palate Infants: A Comparative Evaluation of Cases Done With and Without Nasal Stents</td>
<td>Punga, R., et al.</td>
<td>August 10th /2012</td>
<td>To evaluate cleft lip and cleft palate changes into Two categories</td>
<td>Palatal obturator with unilateral and bilateral molder.</td>
<td>Patient under 2 months of age with a treatment for 9 weeks</td>
<td>It has significant advantages in the treatment of patients with cleft lip and bilateral cleft palate. The improvement of the nostril height was correlated with the time that pre-surgical orthopedics was used.</td>
</tr>
<tr>
<td>Modifications in Presurgical Nasoalveolar Molding Treatment of Bilateral Cleft Lip and Palate Patients With Severely Malpositioned Pre-maxillae</td>
<td>Titiz, S., et al.</td>
<td>2018</td>
<td>To evaluate a modified pre-surgical nasoalveolar molding treatment for patients with bilateral cleft lip and palate with severely misaligned pre-maxilla.</td>
<td>Palatal obturator and Nasoalveolar moulder.</td>
<td>Neonatal patients with a treatment for 5 months</td>
<td>It was proved to get better nose symmetry and it also reduced the cleft significantly.</td>
</tr>
</tbody>
</table>

**Risk of bias**: the articles study type, evidence levels and recommendation degree selected in this review are described: 4 are descriptive, longitudinal and prospective observational studies IIIA, 5 are reports of clinical cases II2 B, 1 is under retrospective study II1 B, 1 is a 1st Cochrane review, 2 are cases and controls IIIB. The level of evidence and grade of recommendation based on the criteria of the Oxford Center for Evidence-Based Medicine (CEBM).

**Results**

Out of the 66,748 articles found in the databases: 4 are descriptive, longitudinal and prospective observational studies, 5 are reports of clinical cases, 1 is a retrospective study, 1 is a Cochrane review, 2 are cases and controls. Articles that were associated with orthopedic treatment with nasal molder and obturator plate were included. On the other hand, papers which did not match the time of use and patients who had undergone some type of oral surgery (Palatorraphy and Sheiloplasty) were excluded.

According to Figure 1, the studies show that the equipment used depended on
the extension of the cleft palate. In case study articles the use of sealing plates were implemented if it was a cleft palatal condition, Nasoalveolar molder if the fissure was labial compromising the base and the nasal ala, or palatal obturator with nasal molder if the fissure of the palatal lip was complete. The initial time of the pre-surgical orthopedics should be from the moment of birth until 3 months of age.

The main functions reported in the studies coincide in the importance of presurgical orthopedics since it manages to seal oral communication, reposition the lingual muscles, stimulates and guides adequate maxillary growth, facilitates feeding and psychologically supports parents in the process they must face.

By sealing the nasal passages and the oral cavity, any irritation of the nasal mucosa is avoided, preventing infections and injuries, improving food conditions and favoring suction, preventing the possibility of bronchoaspiration of liquids or food avoiding them to the respiratory tract. In addition, it balances normal breathing, both intraoral and extraoral air pressure caused during sucking and swallowing.
As for the direct orthopedic action, it prepares the infant for surgery with the approach of the ridges, gives the alveolar ridges a harmonious contour and reduces the deformation of the nasal floor. Also, this process is favored by the participation of the parents in the treatment and the establishment of trusting relationships with the medical team.

Characteristics of the studies: The evidence ranking of the included studies was based on the criteria of the Oxford Center for Evidence-Based Medicine (CEBM). The articles were chosen because they met the eligibility criteria. In addition, the type of study and its level of evidence played an important role. 2 articles belong to I category, 10 more to II-2 category, and the last one (1) to II-3 category type. On the other hand, within the recommendation degree papers 4 articles are located into A type, 7 into B, and 2 into C type.

Discussion

Literature has recognized that patients with cleft lip and cleft palate have negative psychosocial effects due to their esthetics and functional conditions. They may have low self-esteem and quality of life. Therefore, they require a multidisciplinary professional counseling to provide a more individualized treatment.

Millard claimed that the unilateral and bilateral cleft is caused by the mesenchyme failure to migrate from the maxillary prominence to the medial nasal prominence. As a result, the maxilla and pre-maxilla do not form a bony union, either unilaterally or bilaterally, depending on the side / sides of the migration failure; being similar to the studies reported in this review where it was found that the cleft lip and palate were caused by the non-migration of the maxillary, palatal and nasal prominence.

Pre-surgical orthopedics arises as a treatment with good prognosis, based on the principles of McNeil, and Hotz and Gnoinski contributions. The study contributions by Punga R, and Sharma SM are similar to the results found in this review where they show that the pre-surgical orthopedic treatment is carried out by using passive plates to close the palate avoiding lingual interposition and improving feeding. These molders seek the retraction of the pre-maxilla. The nasal molder corrects the nasal deformity using tight tapes force; show that the result is pretty much favorable because changes in the cleft lip, alveolar cleft and nasal morphology were evident in children with nasal moldings.

This type of treatment is ideal to carry out during the neonatal period where high levels of polysaccharide allow the plasticity of the nasal cartilage in the first weeks of the child's life. The alar cartilage could be molded with permanent results if treatment is started within 6 weeks of life. This is based on the fact that during this period there are high levels of estrogen from the mother in the fetal circulation that causes an increase in hyaluronic acid. Hyaluronic acid gives the elasticity of the connective tissue. Estrogen levels begin to drop at 6 weeks of age.

Carrying out this treatment offers an early improvement in regional conditions to reduce the size of the fissure, establish a correct anatomical relationship between the fissured maxillary segments, thus facilitating primary surgery; approaching
of the maxillary segments decreasing the tension of the soft tissue for lip and nose repair; achieve a stable maxillary base for the lip and correct the bone deficiency of the nasal floor; allow stimulation and bone remodeling and weight gain by facilitating feeding in the newborn, among others.

According to the scoping review carried out, the treatment with palatal obturator in patients with cleft lip and palate was from 2 to 4 months using the device 24 hours a day with weekly controls depending on the extension of the fissure. The time of use of the nasoalveolar molder was from 2 to 4 months depending on the nasal deformity, this nasal conformer requires tapes adhered to the cheeks. This scoping review contribution is similar Restrepo NL and collaborators; they claimed that orthopedics is an effective treatment in patients with cleft lip and cleft palate as long as it is performed early to assure favorable results, improving patient’s quality of life. Attiguppe P and collaborators 2016 agree with pre-surgical orthopedics success and benefits if it performed on time. After three months of life, its effectiveness progressively decreases according to the levels of hyaluronic acid. To sum up, the success of orthopedics will depend on if it is used in the early stage of life.

Results have positively influenced parents whose collaboration, help and support are key factors to assure the desire treatment outcomes. According to Duca and collaborators 2013, the importance of incorporating family as mandatory participants will lead to greater support, companion and solidarity which will be a fundamental factor in achieving the objectives.

More studies are needed to obtain proper samples. Taking this into account, future research should consider the performance of high quality randomized controlled trials that assess the effect of pre-surgical orthopedics in patients with cleft lip and palate.

Conclusion

The results of this scoping review agree that presurgical orthopedics is a successful treatment when performed from birth to three months of age.

It offers favorable results by reducing the gap of the palatal cleft, nasal deformity and cleft lip before practicing cheilorrhaphy and palatorrhaphy, which benefits the oral and general health of the child.

With the use of intraoral plates, the maxillary segments are progressively aligned, the premaxilla is retracted and the alveolar edges are approximated; decreasing the probability of the requirement for a secondary alveolar bone graft in mixed dentition, generating stability in nasal and alveolar asymmetry.

Conflicts of interest:

The authors declare that they have no conflicts of interest

Authors’ contributions:

Authorship included participation in study conception, drafting, and final approval of the manuscript. All authors contributed in an articulate and equitable manner.
Referencias bibliográficas


Recibido: 27/11/2020
Aceptado: 19/07/2021
Correspondencia: Cristhian Ariel Cisneros Hidalgo, correo: Cristhian.Cisneros@campusucc.edu.co