Research, Society and Development, v. 11, n. 1, e29111124969, 2022
(CC BY 4.0) | ISSN 2525-3409 | DOI: http://dx.doi.org/10.33448/rsd-v11i1.24969

Secondary alveolar bone graft in incomplete unilateral pre-foramen fissure: case report

Enxerto ósseo alveolar secundário em fissura pré-forame incompleta: relato de caso

Injerto de hueso alveolar secundario en fisura pre-forma incompleta: reporte de un caso

Received: 12/20/2021 | Reviewed: 12/27/2021 | Accept: 12/29/2021| Published: 01/07/2022

Luiza Roberta Bin
ORCID: https://orcid.org/0000-0002-4916-559X
Universidade Estadual do Oeste do Paraná, Brasil
E-mail: luizarbin@gmail.com
Eleonor Álvaro Garbin Júnior
ORCID: https://orcid.org/0000-0002-2111-4766
Universidade Estadual do Oeste do Paraná, Brasil
E-mail: alvarogarbin@yahoo.com.br
Geraldo Luiz Griza
ORCID: https://orcid.org/0000-0001-7169-495X
Universidade Estadual do Oeste do Paraná, Brasil
E-mail: ggriza@hotmail.com
Natasha Magro Érnica
ORCID: https://orcid.org/0000-0002-0545-1623
Universidade Estadual do Oeste do Paraná, Brasil
E-mail: natashamagro@uol.com.br
Mauro Carlos Agner Busato
ORCID: https://orcid.org/0000-0002-8379-9211
Universidade Estadual do Oeste do Paraná, Brasil
E-mail: mcabusato@uol.com.br
Ricardo Augusto Conci
ORCID: https://orcid.org/0000-0002-6678-8780
Universidade Estadual do Oeste do Paraná, Brasil
E-mail: ricardo_conci@hotmail.com

Abstract
Cleft lip and palate is one of the most common facial deformities. During embryonic life, non-fusion of the maxillary and medial nasal plaques leads to cleft lip and palate. Fissures can produce a range of dental problems in terms of number, size, shape, and position, related to deciduous or permanent dentition. Besides this, the teeth most affected are those located in the fissure area. There are numerous treatment protocols, which, despite the lack of a consensus, start as soon as the child is born, going into adulthood, seeking functional and aesthetic rehabilitation. The surgical phases, lip repair, nasal repair, palatoplasty and alveolar bone grafting, are performed according to age. As for the bone graft, the most used option is the secondary graft, with the autogenous one being the most available. Thus, the objective of this work is to present a clinical case of secondary alveolar bone grafting in a 10-year-old female patient with an incomplete unilateral pre-foramen cleft.
Keywords: Cleft palate; Alveolar bone grafting; Orthodontics.

Resumo
A fissura labiopalatina é uma das deformidades faciais mais comuns. Durante a vida embrionária, a não fusão das placas nasais maxilar e medial leva à fenda labial e palatina. As fissuras podem produzir uma série de problemas dentários em termos de número, tamanho, forma e posição, relacionados à dentição decídua ou permanente. Além disso, os dentes mais afetados são aqueles localizados na área da fissura. São inúmeros os protocolos de tratamento, que, apesar da falta de consenso, se iniciam desde o nascimento da criança, chegando à idade adulta, em busca da reabilitação funcional e estética. As fases cirúrgicas, reparo labial, reparo nasal, palatoplastia e enxerto ósseo alveolar, são realizados de acordo com a idade. Quanto ao enxerto ósseo, a opção mais utilizada é o enxerto secundário, sendo o autógeno o mais disponível. Assim, o objetivo deste trabalho é apresentar um caso clínico de enxerto ósseo alveolar secundário em uma paciente de 10 anos de idade com fissura pré-forame unilateral incompleta.
Palavras-chave: Fenda palatina; Enxerto ósseo alveolar; Ortodontia.

Resumen
El labio leporino y el paladar hendido es una de las deformidades faciales más comunes. Durante la vida embrionaria, la no fusión de las placas nasales maxilar y medial conduce a labio leporino y paladar hendido. Las fisuras pueden producir una serie de problemas dentales en cuanto a número, tamaño, forma y posición, relacionados con la dentición temporal o permanente. Además, los dientes más afectados son los que se encuentran en la zona de la fisura. Existen numerosos protocolos de tratamiento, que, a pesar de la falta de consenso, parten del nacimiento del niño, llegando a la edad adulta, en busca de una rehabilitación funcional y estética. Las fases quirúrgicas, reparación de labios, reparación nasal, palatoplastia e injerto de hueso alveolar, se realizan según la edad. En cuanto al injerto óseo, la opción más utilizada es el injerto secundario, siendo el autógeno el más disponible. Así, el objetivo de este trabajo es presentar un caso clínico de injerto de hueso alveolar secundario en un paciente de 10 años con hendidura preforamen unilateral incompleta.
Palabras clave: Paladar hendido; Injerto de hueso alveolar; Ortodoncia.

1. Introduction

Cleft lip and palate is one of the most common facial malformations (Alyami et al. 2020; Huang, Chen, PhD, and Chen, PhD 2021; Mituuti et al. 2010). Epidemiologically, the literature reports variable numbers, such as 1 affected individual in every 700 individuals (Farronato et al. 2014). Approximately 80% of these anomalies are not syndromic in origin (Kaura et al., 2018). This anomaly is variable and may comprise lip only, lip and palate, or palate only (Mituuti et al. 2010). The etiology of the fissures is complex and multifactorial, and the genetic and environmental influence on the critical stages of embryonic development is well known (Farronato et al. 2014). Disorders in the fusion process of the maxillary and nasal processes may lead to fissures (Huang et al., 2021; Mituuti et al. 2010). There are a number produce a range of dental problems in terms of number, size, shape, and position, involving deciduous or permanent dentition (Farronato et al. 2014; Huang et al., 2021).

These anomalies lead to multiple sequelae, requiring multidisciplinary treatment (Farronato et al. 2014; Kaura et al., 2018). This has been discussed without a consensus, with numerous treatment protocols that have evolved, starting as soon as the child is born and proceeding into adulthood, with the aim of achieving functional and aesthetic rehabilitation (Farronato et al. 2014). Appropriate treatment depends on a complete group of professionals (Farronato et al. 2014; Kassam et al. 2020; Torres et al., 2019). Particularly, surgical repair is important for the preservation of facial growth, normal speech formation, and adequate dental development (Farronato et al. 2014; Kassam et al. 2020). It is also important to note that the smallest possible number of surgeries is ideal, in order to ensure less scarring and avoid growth delay (Farronato et al. 2014). The surgical phases are divided according to age (Farronato et al. 2014).

Cheiloplasty and palatoplasty vary in techniques and timing (Farronato et al. 2014). As for the alveolar bone graft, the technique has been discussed since 1900, but it was only after the middle of the century that the procedure became disseminated (Farronato et al. 2014). Questions arise mainly regarding the type and timing of the performance (Farronato et al. 2014; Mahajan et al. 2017; Torres et al., 2019). This treatment can be classified as primary, performed after cheiloplasty but before palatoplasty; secondary, performed during mixed dentition; and tertiary, performed after the complete development of permanent dentition (Kaura et al., 2018). The secondary graft has been the most used (Kaura et al., 2018; Mahajan et al. 2017), as it offers a higher rate of success in gaining periodontal insertion to adjacent teeth, allowing canine eruption and dental alignment, while decreasing the facial growth restriction (Mahajan et al. 2017). The options for the graft are numerous (Ralph Millard 1980), the main one being autogenous bone, with the source being only one of the variables for success (Kaura et al., 2018). Among them, chondral cost, iliac crest, tibia and vomer can be mentioned (Ralph Millard 1980). For greater predictability, the evaluation of the bone remains is necessary, with radiographs being used for this purpose (Kaura et al., 2018; Mahajan et al. 2017), while bone quantity is classified according to the Enemark scale (Enemark et al., 1987; Mahajan et al. 2017). It varies in levels I, II, III, and IV, where 75%, 50-75%, 25-50%, and 25% of the alveolar bone height are observed (Enemark et al., 1987; Mahajan et al. 2017).

The purpose of this work is to present a clinical case of treatment of a patient with incomplete unilateral pre-foramen

cleft, highlighting the surgical phase of the secondary alveolar bone graft.

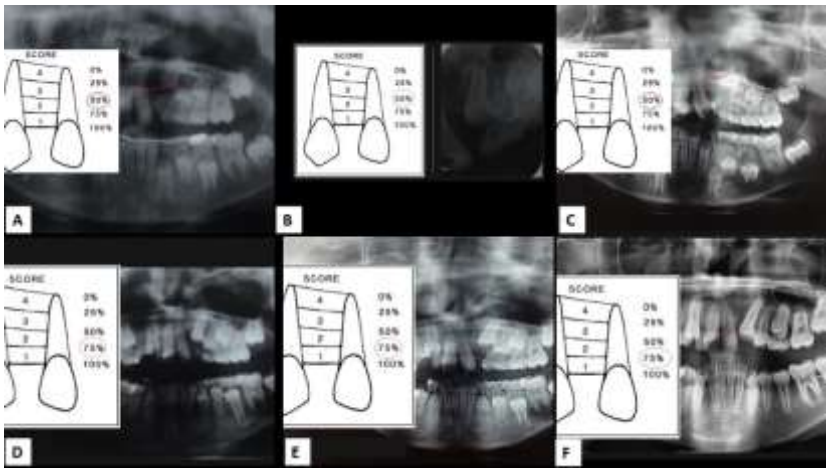
2. Case Report

At the Care and Research Center for Craniofacial Anomalies (CEAPAC/HUOP/UNIOESTE), an 8-year-old female patient who had an incomplete left unilateral pre-foramen cleft was treated by the Oral and Maxillofacial Surgery team. On clinical examination, a vertical scar was observed on the upper left lip, with mixed dentition gyroversion of permanent upper central left incisor at almost 180°, enamel hypoplasia in the deciduous upper left canine, and no changes in the palate (Figures 1A, B, and C). In the radiographic evaluation, the tooth germ of the upper lateral left incisor was absent, the bone remnant in the fissure was classified as Enemark level III, and the root of the upper left canine was observed with about 1/3 of the root formed (Figure 2A). The treatment proposed encompassed performing a bone graft in the region to make orthodontic treatment feasible and to provide bone conditions for the eruption of the permanent maxillary left canine. Previously, the patient had undergone cheiloplasty at 7 months, with maxillary expansion using a Hyrax device. Additionally, at the time of the alveolar bone graft phase, at 10 years old, she had a transpalatal arch implemented, having already undergone reanatomization of permanent upper central left incisor and extraction of the deciduous upper left canine (Figures 1D, E, and F). New radiographs were taken, in which the remained bone was classified as Enemark level III, and the root of the upper left canine was observed with about 2/3 of the root formed (Figures 2B and C). The bone graft of choice was autogenous bone, and the donor area was the iliac crest. The procedure was performed under general anesthesia. Removal was performed by the HUOP orthopedics team (Figure 3A). After a rectilinear intrasulcular incision from the left upper central incisor to the left upper first molar, with anteroposterior oblique incision in its center (Figure 3B), with subsequent mucoperiosteal detachment and fistula suture on the nasal floor (Figure 3C). The block was particulate, being accommodated in the fissure (Figure 3D), and the suture was performed with Monocryl (Figure 3E). There were no trans-operative complications. In the postoperative evaluations, tissues were observed in normal repair process (Figure 3F). In the postoperative radiographic evaluation, a satisfactory bone level gain was observed (Figure 2D). On the 2-month postoperative radiograph, the Enemark level was classified as grade II (Figure 2E). Permanent upper left canine began its eruption (Figure 1E and F), with poor dental positioning on the right side (Figure 1D), maintaining the bone level (Figure 2F) and was orthodontically tractioned at the location of the fissure (Figure 1G and H) . Orthodontic treatment is ongoing (Figures 1G and H), requiring follow-up until bone growth of the face is complete. Follow-up by the OMFS team continues and, in the event that it is necessary to correct the maxillomandibular discrepancy, orthodontic preparation and orthognathic surgery will be performed. Written informed consent for patient information and images to be published was obtained from the patient's father.

Figure 1. 1a, 1b, 1c: Initial intraoral clinical aspect; 1d, 1e, 1f: Clinical appearance after 2 years and 2 months postoperatively; 1g and 1h: Clinical appearance after 3 years and 5 months postoperatively.

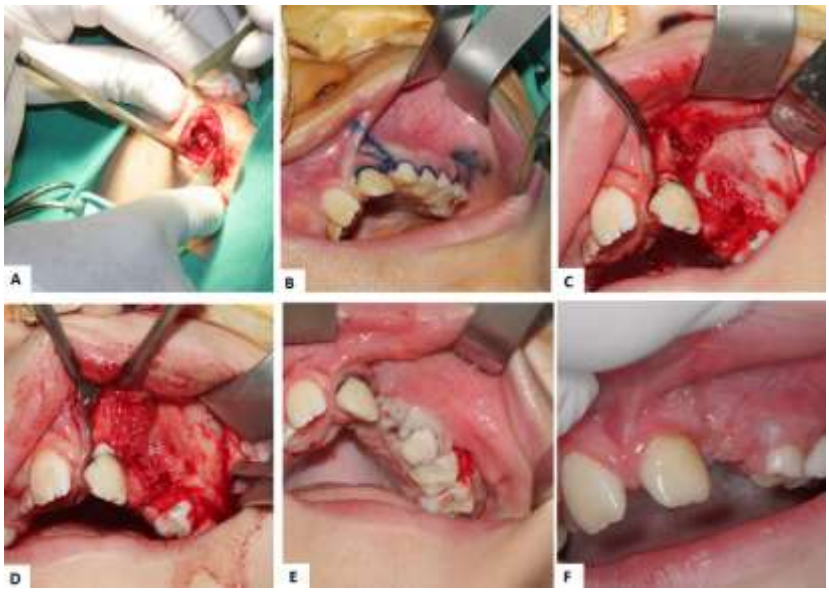
Fonte: Autores.

Figure 2. Radiographic evaluation and classification of the bone remnant according to the Enemark scale in: 2a:Initial; 2b and 2c. After maxillary expansion and preoperatively; 2d: After secondary bone graft. 2e: After 6 months of the secondary bone graft; 2f: After 2 years and 2 months of the secondary bone graft.



Fonte: Autores.

Figure 3. 3a: Collection of bone graft from the donor area of the iliac crest. 3b: Marking of the incision; 3c: Mucoperiosteal detachment and suture of the nasal mucosa to close the fistula. 3d: Accommodation of the autologous bone graft with iliac crest. 3e: Suture in position with Monocryl. 3f: Clinical appearance after 28 days postoperatively.



Fonte: Autores.

3. Discussion

The alveolar bone graft aims to restore function and aesthetics to the patient, offering a support area for teeth eruption, joining the maxillary arch segments and improving lip support and facial aesthetics (Mahajan et al. 2017; Scalzone et al. 2019; Vellone et al. 2017), correction of oronasal fistulas, and support to the alar base and adjacent soft structures (Desai et al. 2019; Lowry et al. 2021; Mahajan et al. 2017), in addition to support for dental eruption and for the periodontium, establishing the nasal skeletal base, nose floor and functional airway, providing bone for the future installation of a dental implant in case of lateral incisor agenesis (Doucet et al. 2019; Mahajan et al. 2017).

The best time to perform the graft is widely discussed (Bittermann et al., 2018; Coots 2012; Enemark et al., 1987; Kaura et al., 2018; Sancak et al. 2019). In general, surgery should be performed on the mixed dentition, prior to the eruption of the permanent canine or before the lateral incisor, when present (Coots 2012; Kaura et al., 2018), being generally performed between 7 and 12 years of age (Coots 2012; Mahajan et al. 2017; Sancak et al. 2019). The literature shows that it is ideal to implement the graft when the root of the canine or lateral incisor has at least 1/4 to 1/3 of its root formed, when it would be the active period of the eruption (Brudnicki et al. 2020; Mahajan et al. 2017). In this case, the patient underwent bone graft at 10 years of age, in mixed dentition and with about 2/3 of the canine root formed. The delay in carrying out the graft may compromise the periodontium of the upper central incisor or lateral incisor on the side of the fissure, as well as disproportion between crown and root, which can limit orthodontic and rehabilitation treatment (Doucet et al. 2019; Mahajan et al. 2017).

Autogenous bone is considered the gold standard for bone grafting (Desai et al. 2019; Kaura et al., 2018; Scalzone et al. 2019), as it offers osteogenic cells, which is an essential factor for bone regeneration and neoformation (Desai et al. 2019; Scalzone et al. 2019). It provides scaffold for osteoconduction, growth factors for osteoinduction, and progenitor cell for osteogenesis (Desai et al. 2019). This The autogenous bone graft is potentially viable, as in ideal conditions, osteogenic cells survive the surgical procedure, particularly in spinal graft (Bergland et al., 1986). Cell survival is highly related to access to nutrients during the critical ischemic phase immediately after graft insertion (Bergland et al., 1986). For medullary bone,

revascularization begins at around 5-8 days, with the graft being completely revascularized at 21 days (Bergland et al., 1986). The cortical bone takes a longer time, as it undergoes the remodeling process with new formation, resulting in new haversian systems, and the grafted bone is completely replaced by a new one (Coots 2012). Conversely, traumatic surgeries can result in grafts that are unable to revascularize (Bergland et al., 1986). Furthermore, alveolar defects should be evaluated as pyramids, considering the importance of filling the bases with cortical bone, in order to provide greater resistance to the natural contraction of tissues during the repair process, maintaining the volume and potentially reducing the need for new grafts (Coots 2012).

Additionally, the choice of donor area depends on factors such as the surgeon's experience, the volume of bone required, and the morbidity of the collection area (Scalzone et al. 2019), using mainly the iliac crest, mandible, or tibia (Desai et al. 2019; Scalzone et al. 2019). The iliac crest is the most widely chosen source thanks to its osteogenic properties, its spinal part being the most often used alongside the cortical part, to greater provide resistance (Coots 2012; Desai et al. 2019). Its disadvantages, in particular, may include scarring, pain, impaired walking, potential nerve damage (Coots 2012; Mehta et al. 2018), and subsequent long periods of hospitalization (Sancak et al. 2019; Scalzone et al. 2019). Therefore, the choice of autogenous bone graft from the iliac crest donor area is justified by the properties it presents and its availability.

Studies have revealed graft alternatives to reduce morbidity (Coots 2012; Mehta et al. 2018; Scalzone et al. 2019) and surgical time, although, for some authors, surgical time is not considered a disadvantage of the autogenous bone graft (Scalzone et al. 2019). The most widely used alternative, in addition to autogenous bone, is recombinant human bone morphogenetic protein-2 (rhBMP-2), which has an osteogenic function, as well as being autogenous, with the greatest advantage of eliminating the morbidity of the donor area (Scalzone et al. 2019). Despite this major advantage, a systematic review performed comparing the autogenous graft with rhBMP-2 in cleft lip and palate patients undergoing secondary bone graft and observed that the autogenous was better in the first 6 postoperative months, but after 1 year there were no differences in height or bone volume (Scalzone et al. 2019). The disadvantages of this material include its high cost and limited availability (Coots 2012). Nevertheless, Mehta et al. (2018) showed in a study that the costs for the hospital are not higher, as only one team is needed for the procedure and the hospital stay is shorter – factors that would decrease costs and subsequently make the rhBMP-2 accessible (Mehta et al. 2018).

Preoperative orthodontics is essential and highly recommended (Kaura et al., 2018; Ralph Millard 1980; Torres et al., 2019; Vellone et al. 2017). Prior expansion has the role of expanding the segments, in order to facilitate access to the fissure and ensure space for the graft (Kaura et al., 2018; Ralph Millard 1980). Ideally, the expansion should be performed at least 6 months before surgery (Kaura et al., 2018). In this case, the patient underwent pre-grafting expansion. After surgery, orthodontics must continue in order to restore the symmetry of the arch when permanent dentition is achieved, closing the space or providing conditions for the installation of osseointegrated implants, both helping to reduce bone resorption (Kaura et al., 2018). A bone bridge with sufficient height and width is important for the success of the graft and to guide the eruption or orthodontic movement of the permanent tooth in the region (Mahajan et al. 2017). It was observed, therefore, that the orthodontic management for this case was consistent with that recommended in the literature. Where necessary, pre-orthognathic orthodontics must decompensate to offer the best intercuspation possible, leading to greater stability (Farronato et al. 2014; Ralph Millard 1980). Moreover, postoperative orthodontics should refine the tooth movements still needed (Farronato et al. 2014; Ralph Millard 1980). Thus, the patient is undergoing orthodontic treatment, aligning the teeth in their arches, and is expected undergo a new evaluation at the end of the growth phase.

The success of the secondary bone graft depends on numerous parameters, such as presence of a deciduous tooth around the fissured area, gingival health, presence of canine or lateral incisor, position of the premaxilla, bone quality around the fissured area and related to the present tooth, postoperative complications, and need for a surgical reapproach (Mahajan et al. 2017;

Scalzone et al. 2019). Enemark's scale of radiographic assessment was applied to verify the high of the bone on cleft region and it was found the gain and maintenance of the level of the bone in this case. The success of the graft is achieved with oral rehabilitation, determined by the eruption of permanent dentition, closure of the oronasal fistula, and lip and alar-based support (Kaura, Srinivasa, and Kasten 2018; Scalzone et al. 2019). Dental eruption in the grafted region can happen only after the presence of vital bone and evidence of bone transformation (Kaura et al., 2018). Although clinical evaluation is one of the bases for measuring the success of the procedure, it is also necessary to check the patient's quality of life, as the improvement in self-esteem leads to better socialization and acceptance (Ralph Millard 1980; Sancak et al. 2019). Conversely, dissatisfaction in the treatment can lead to negative effects on the individual's personal and professional life (Ralph Millard 1980; Sancak et al. 2019). According to radiographic and clinical evaluation, it can be said that this case is successful in the alveolar bone graft, as it was possible to traction the tooth in the cleft region, restoring function and aesthetics to the patient. Additionally, the patient's guardian reported an improvement in social interaction and greater school productivity, suggesting an improvement in quality of life.

4. Conclusion

Due to the complexity of anomalies in the development of cleft lip and palate patients, multidisciplinary treatment is essential. It should start after birth, requiring follow-up until adulthood. Secondary alveolar bone graft is indispensable in the integrated treatment of these patients, and its success depends mainly on its implementation in the ideal period, when the root of the canine has reached up to 2/3 formation.

Acknowledgments

The authors would like to acknowledge each professional of the medical team, laboratory, and imaging center, that was involved in each phase of the treatment of the patient but do not fulfill the authorship criteria.

References

Alyami, B. et al. (2020). "Prevalence and Clinical Case Series of Syndromic and Nonsyndromic Cleft Lip and Palate in a Saudi Arabian Neonatal Population." *Cleft Palate-Craniofacial Journal* 57(11): 1259–65.

Bergland, O., Gunvor, S., & Frank, E. A. (1986). "Elimination of the Residual Alveolar Cleft by Secondary Bone Grafting and Subsequent Orthodontic Treatment." *Cleft Palate Journal* 23(3): 1–22.

Bittermann, Arnold J. N, Aebele B Mink Van De Molen, & Robert Jj. (2018). "Midfacial Growth and Dental Arch Relationships in Bilateral Cleft Palate Following Secondary Alveolar Bone Grafting and Orthodontic Intervention: Factors Predicting a Le Fort I Osteotomy at Age 18." *Journal of Cranio-Maxillo-Facial Surgery* 46(10): 1764–71.

Brudnicki, A., Ewa Sawicka, R. B., & Piotr S. F. (2020). "Effects of Different Timing of Alveolar Bone Graft on Craniofacial Morphology in Unilateral Cleft Lip and Palate." *Cleft Palate-Craniofacial* Journal 57(1): 105–13.

Coots, Br. K. (20120. "Alveolar Bone Grafting: Past, Present, and New Horizons." *Seminars in Plastic Surgery* 26(4): 178–83.

Desai, A. K., et al. (2019). "Efficacy of Platelet-Rich Fibrin in Secondary Cleft Alveolar Bone Grafting." *Craniomaxillofac Trauma Reconstruction Open* 3(1): e43–50.

Doucet, J C., et al. (2019). "Facial Growth of Patients With Complete Unilateral Cleft Lip and Palate Treated With Alveolar Bone Grafting at 6 Years." *Cleft Palate-Craniofacial Journal* 56(5): 619–27.

Enemark, H., Steen S.-P., & Mette B. (1987). "Long-Term Results after Secondary Bone Grafting of Alveolar Clefts." *J Oral Maxillofac Surg* 45(1): 913–18.

Farronato, G., et al. (2014). "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." *Stomatologija* 16(2): 53–60.

Huang, S., Yang C., and Zhenqi C. (2021). "Relationship Between Crown to Root Ratio and Alveolar Bone Support in Patients With Unilateral Cleft Lip and Alveolus." *Cleft Palate-Craniofacial Journal* 58(2): 215–21.

Kassam, S. N. et al. (2020). “World Cleft Coalition International Treatment Program Standards.” *Cleft Palate-Craniofacial Journal* 57(10): 1171–81.

Kaura, A. S., Dhivya R. S., & Steven J. K. (2018). “Optimal Timing of Alveolar Cleft Bone Grafting for Maxillary Clefts in the Cleft Palate Population.” *Journal of Craniofacial Surgery* 29(6): 1551–57.

Lowry, C. H. et al. (2021). “The Effect of Earlier Bone Grafting, Prior to Orthodontic Treatment, on SWAG Ratings of Graft Outcomes.” *Cleft Palate-Craniofacial Journal* 58(2): 208–14.

Mahajan, R. et al. (2017). “Evaluation of Secondary and Late Secondary Alveolar Bone Grafting on 66 Unilateral Cleft Lip and Palate Patients.” *Plastic Surgery* 25(3): 194–99.

Mehta, S. et al. (2018). “Cost-Effectiveness Analysis of Demineralized Bone Matrix and RhBMP-2 versus Autologous Iliac Crest Bone Grafting in Alveolar Cleft Patients.” *Plastic and Reconstructive Surgery* 142(3): 737–43.

Mituuti, C. T., Silvia H. A. P-P, Giovana R. B., & Cristina G. de A. B-G. (2010). “Caracterização Da Fala de Indivíduos Submetidos à Palatoplastia Primária.” *Revista da Sociedade Brasileira de Fonoaudiologia* 15(3): 355–61.

Ralph Millard. (1980). III Plastic and Reconstructive Surgery Cleft Craft: The Evolution of Its Surgery—Volume III: Alveolar and Palatal Deformities.

Sancak, K, et al. (2019). “Effect of Alveolar Bone Grafting on Health Quality in Patients With Cleft Lip and Palate.” *The Journal of craniofacial surgery* 30(8): e771–74.

Scalzone, A., et al. (2019). “Secondary Alveolar Bone Grafting Using Autologous versus Alloplastic Material in the Treatment of Cleft Lip and Palate Patients: Systematic Review and Meta-Analysis.” *Progress in Orthodontics* 20(1).

Torres, A., et al. (2019). “A Programmable Expander for Patients with Cleft Lip and Palate.” *Cleft Palate-Craniofacial Journal* 56(6): 837–44.

Vellone, V., et al. (2017) “Canine Eruption after Secondary Alveolar Bone Graft in Unilateral Cleft Lip and Palate Patients.” *Journal of Craniofacial Surgery* 28(5): 1206–10.