The Relation between Maxillary Sinuses and Cleft Lip and Palate

A Relação Entre os Seios Maxilares e a Fissura Labiopalatina

Carolina Silvano Vilarinho da Silva^{ab}; Natalia Garcia Santaella^c; Mariana da Silva Bonatto^d; Luiz Evaristo Ricci Volpato^c; Andreza Maria Fábio Aranha^c; Alexandre Meireles Borba^{*ab}

^aUniversidade de Cuiabá, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery. MT, Brazil.
^bHospital Geral de Cuiabá, Research Program on Integrated Dental Sciences. MT, Brazil.
^cUniversidade de Cuiabá, Faculty of Dentistry, Research Program on Integrated Dental Sciences. MT, Brazil
^dHospital Geral de Cuiaba, Department of Oral and Maxillofacial Surgery, General, Brazil,
*E-mail: Aleborba80@hotmail.com

Abstract

Cleft lip and palate are anomalies of craniofacial development that require reparative surgery to provide considerable improvement in the individual's quality of life, but which can also have a negative impact on the maxilla development. The aim of this study was to understand the influence of surgical repair of cleft lip and palate on the volume of the maxillary sinus of these individuals. A narrative review was carried out using the scientific databases Pubmed and Scielo, where the articles selected by the abstract were fully read, categorized and critically analyzed. There are reports of high frequency of chronic sinusitis in patients with cleft lip and palate, especially children, that may be related to hypoplasia of the maxilla and lower volume of the maxillary sinus related to the cleft. In addition, these individuals are submitted to several surgical corrections that can also negatively influence the maxilla development due to tissue fibrosis, with suggestions for surgical techniques to reduce this interference. Thus, the patient with cleft lip and palate may present maxillary underdevelopment, which may result in hypoplasia of the maxillary sinus, and reparative surgeries can aggravate this complication. This review suggests some surgical techniques to reduce this interference, although there are few articles with clinical trials found in the literature that can explore these alternatives and correlate the results with current surgical protocols.

Keywords: Cleft Palate. Tomography. Sinusitis. Review.

Resumo

As fissuras labiopalatinas são anomalias de desenvolvimento craniofacial que necessitam de cirurgias reparadoras para proporcionar melhora considerável à qualidade de vida do indivíduo, mas que também podem repercutir negativamente no desenvolvimento da maxila. O objetivo deste estudo foi compreender a influência das cirurgias reparadoras de fissuras labiopalatais no volume do seio maxilar desses indivíduos. Foi realizada uma revisão narrativa pelas bases de dados científicos Pubmed e Scielo, de onde os artigos selecionados pelo abstract foram lidos na íntegra, categorizados e analisados criticamente. Há relatos de alta frequência de sinusite crônica em pacientes com fissuras labiopalatais, principalmente crianças, que podem estar relacionada a hipoplasia de maxila e menor volume do seio maxilar relacionado à fissura. Além disso, esses indivíduos são submetidos a diversas correções cirúrgicas que também podem influenciar negativamente o desenvolvimento da maxila devido a fibrose tecidual, havendo sugestões de técnicas cirúrgicas para diminuir essa interferência. Então, o paciente com fissuras labiopalatais pode apresentar hipodesenvolvimento maxilar podendo ter como consequência a hipoplasia do seio maxilar e as cirurgias reparadoras podem agravar essa complicação. Essa revisão sugere algumas técnicas cirúrgicas para diminuir essa interferência, embora existam poucos artigos com ensaios clínicos encontrados na literatura que possam explorar essas alternativas e correlacionar os resultados com os protocolos cirúrgicos atuais.

Palavras-chave: Fissura Palatina. Tomografia Computadorizada. Seio Maxilar.

1 Introduction

Cleft lip and palate (CLP) consist of a common anomaly of craniofacial development, resulting from failure in the fusion of face processes during fetal development¹. Cleft lip (CL) results from non-fusion or defective fusion of the median nasal process with the maxillary process, while cleft palate (CP) is due to the fusion failure of the palatine crests. The estimated occurrence of lip and/or palate clefts in Brazil is 1 to 650 births, representing the most common facial features². The occurrence of CL with CP is frequent, around 45% of the cases. Individuals with CLP present numerous problems regarding nutrition, speech, hearing, chronic infections of the upper airways, dentition, face morphology and psychological aspects³, and thus they need multiprofessional care and repair surgeries of lip, palate, dental alveoli, nose and facial skeleton.

Because they present alterations in the facial development, individuals with CLP may have the morphologically affected maxillary sinus (MS), since it is also originated from the structures of the first pharyngeal arch and occupies an important part of the maxilla body¹. Studies on the development of MS in cases of FLP indicate that the presence of cleft may be related to sinusitis^{4,5}. Individuals with cleft present a high incidence of maxillary sinusitis⁶, however, its etiology is not fully understood^{3,7} and there are controversial opinions. The main emphasized factors are morphological alterations of MS, maxillary sinus floor ascension, nasal obstruction, deviated septum, impaired mucociliary, increased maxillary sinus floor height and nasal cavity lateral wall abnormalities^{3,7}. Another aspect that explains why sinusitis is more frequent in individuals with CLP is that, if these individuals present maxilla hypoplasia, they will have a hypoplastic SM. The hypoplastic sinus will cause drainage pathology due to poor ostium positioning and make a predisposition to sinusitis³.

Thus, the assessment of the maxillary sinus volume (MSV) of individuals with CLP may be useful in determining possible relation to the presence of sinusal diseases (3). Thus, the objective of this review is to understand the influence of FLP repair surgeries on maxillary sinus volume from a narrative review.

2 Development

2.1 Methodology

The present study is a narrative review, whose material collection process was performed in a non-systematic manner from April to May 2020. The scientific databases used for the research were Pubmed and SciELO, from which the selected articles were read in full, categorized and critically analyzed.

2.2 Results and Discussion

2.2.1 Maxillary Sinuses

According to Som and Curtin⁸, the maxillary sinuses are formed from projections of the nasal cavities and increase in size with age. Another study reports that at birth its measurements are approximately 1cm³ and at adulthood 15cm^{3 9}

The functions proposed for the maxillary sinus are air conditioning, skull damping in case of trauma, reduction of skull weight, water buoyancy, increase of the olfactory area, mechanical rigidity, vocal resonance, reduction of the auditory feedback, mucosal production and bone strengthening.^{8,10,11}

2.2.1 Maxillary sinuses and lip and palatine clefts

In a patient with unilateral cleft lip and palate, the maxilla hypoplasia affects the height and not the width of the maxillary sinus being more affected the side without the cleft than with the cleft and Eckel¹² believes that hypoplasia occurs more in the ventral region of the antrum (alveolar process).

Some authors have observed that the development of the maxillary sinus was equal for both the lip and palatine cleft group and the control group, reporting a rapid growth of the maxillary sinus at ages 1 to 8 years, which decreases from 8 to 12 years, representing the stop of growth that approaches the maturation of the antrum. They also concluded that lip and palatine cleft has little or no effect on the size, shape or rate of development of the maxillary sinus since its study did not show significant anatomical difference between the lip and palate cleft and control groups.⁹

Considering that sinusitis is a common problem in individuals with lip and palate cleft, not yet fully understood,

many studies have been directed to try to unravel its origin, which may be related to nasal airway obstruction, deviated septum, increased maxillary sinus floor height, nasal cavity lateral wall abnormalities, reflux, velopharyngeal insufficiency, presence of pharyngeal flap, nasal mucociliary function impairment, oral maxillary sinus contamination, altered air flow through the nose and sinuses of the face, food problems, secretion aggravated by velopharyngeal insufficiency or fistula and the poor positioning of the ostium in a deficient maxillary sinus that leads to pathological drainage and then to sinusitis predisposition, but none of these factors were definitively indicated as the cause of sinusitis.^{5,13-16} Studies carried out in individuals without lip and palatine cleft question the implication of concha bullosa, deviated septum or decrease in nasal airflow secretion as factors for rhinitis and sinusitis¹⁷⁻¹⁹

By means of a study in which the relation between middle ear and sinusitis pathologies in Navajos Indian children was evaluated, the authors reached some possible explanations for sinusitis, such as saliva reflux and food with irritation of the sinus mucosa, maldevelopment of the maxilla and palate that can present a small, partial or totally obliterated ostium with a poor position or displaced laterally secondary to a deviated septum. Anatomically, this may be explained by the septum deviated to the side without cleft and unilateral and straight lip and palate cleft in median cleft.¹⁶

The velopharyngeal incompetence in patients with lip and palate cleft may lead to regurgitation of food and saliva with edema and irritation of the nasal mucosa and maxillary sinus and obliterates the natural ostium resulting in sinusitis, but with the need for further studies in order to have this confirmation.⁹

Some studies have determined that the most common anomaly in patients with chronic sinusitis is the hypoplasia of the maxillary sinus that is still accompanied by lateral nasal wall abnormality justified by chronic inflammation in the active maxilla to osteogenesis promoting changes in maxillary sinus pneumatization, which decreases its volume, justifying this in individuals with lip and palatine cleft, who present previous episodes of chronic sinusitis.^{20,21}

Whereas in some studies comparing the maxillary sinus volume of individuals with unilateral, bilateral lip and palatine cleft and control group without clefts through the analysis of computed tomography of the conical beam and concluded that the maxillary sinus volume was lower in individuals with unilateral lip and palatine cleft compared to the bilateral without finding statistically significant difference between the right and left sides of the groups, others found the maxillary sinus larger on the non-clefted side in relation to the clefted side of the unilateral lip and palatine cleft although with little statistical significance.^{11,22-24} Some authors also concluded that sinusitis was more severe on the non-clefted side of the patient with unilateral lip ad palatine cleft.^{15,22}

Studies comparing the maxillary sinus volume of

individuals with unilateral lip and palatine cleft and individuals without cleft as a control group through the analysis of conic beam computed tomography have identified that the mean maxillary sinus volume of individuals with lip and palatine cleft was lower than that of the control group with a statistically significant difference, which can be justified by the fact that lip and palatine cleft may affect the maxillary sinus volume because in these individuals the maxillary sinus develops differently in the embryonic period.^{3,7,11,22} However, this finding is in conflict with studies whose analysis of the maxillary sinuses used two-dimensional evaluation. It is known that the lateral cephalometric X-ray used for two-dimensional analysis is simple and low cost but has limitations such as distortion, low resolution due to the difficulty identifying the reference points, differences in magnification and overlap of craniofacial structures, and that computed tomography of the conical beam allows greater precision in the three-dimensional evaluation of the sinus beyond the low radiation dose and shorter time for image acquisition. 22,25-27

A study comparing the amount of mucosal thickening and the size of the maxillary sinus in children with lip and palatine cleft and without cleft pairing age and sex by means of a comparative tomographic analysis, concluded that the presence or absence of lip and palatine cleft significantly affects all maxillary sinus measurements and also stated that a child with cleft tends to have the maxillary sinus smaller than a child without cleft.^{9,11} Some authors report that mucosal thickening may be related to periapical and periodontitis lesions, especially in adolescents and elderly individuals, and that sinusitis in children is rarely related to dental infections, therefore, a future study could evaluate the relation of thickening with dental disease and children with lip and palatine cleft²⁸⁻³¹

In the study that compared the adult skull with lip and palatine cleft with 140 skulls of individuals without cleft suggested a close connection between the size of the facial skeleton and the maxillary sinus volume even before a severe facial pathology, however, the analysis of only one skull with lip and palatine cleft limited its study in addition to the fact that most of these individuals are surgically treated and cannot say how the condition can affect the maxillary sinus volume.⁶ Although some authors have suggested that the individual with unilateral lip and palatine cleft has hypoplastic jaws if the maxillary sinus size reflects the maxilla size.³²

The lip and palatine cleft teams should be aware that children with unilateral lip and palatine cleft may have significant thickening of the sinus mucosa, because according to their study these children presented thickening 3 times greater than that of the control group, and this must be examined to determine its cause and the need for treatment.¹¹

Surgical corrections may negatively influence the maxilla development probably due to tissue fibrosis. Some authors who compared two surgical protocols of palatoplasty to evaluate craniofacial morphology, being that one protocol would be the surgical approach that divides palatoplasty into two surgical times and the other protocol would be palatoplasty performed in a single time, they concluded that the single-time approach had better results with a repair that contributes little to the retro positioning of the maxilla regardless of the surgical technique used even if this closure was delayed to 4 or 5 years of age.³³⁻³⁷ Although other studies state that all types of palatoplasty generate maxilla transverse hypoplasia and with a palatoplasty divided in to two times trend have a lower rate of palate narrowing and cross-bite , but there is a need for further analysis for this statement.³⁸⁻⁴⁰

3 Conclusion

Lip and palate cleft presents a complex anatomy that must be considered for the treatment of this deformity to be complete, as well as wide and coordinated care from childhood and throughout adolescence, since the balance between intervention and growth is essential, in addition to the good training and experience of the professionals involved in their treatment, being centered on the patient, family and community.

No studies were found in this review comparing surgical repairs of cheiloplasty, palatoplasty and alveolar bone grafting in different age groups with operated and non-operated cases to analyze craniofacial growth of individuals with lip and palatine cleft, since the maxilla development is directly related to the growth of the maxillary sinus, no studies were found, correlating these surgical repair procedures with the maxillary sinus volume.

It is worth pointing out that the patient with lip and palatine cleft may present maxillary hypo development and consequently maxillary sinus hypoplasia, which may result in sinusitis in these individuals, and the surgical procedures for repairing these clefts may further aggravate this condition, in addition to the recurrent sinusitis profile may be a complicating factor for the success of alveolar bone graft surgeries, with the possibility of a reduction in quality of life. Therefore, the surgical team responsible for these repairs must have the knowledge of these complications and always seek for surgical techniques and adequate moments in order to minimize them.

There is a need for further research that analyzes each surgical repair of lip and palatine cleft and its potential for interference of craniofacial development so that the limitations of surgical techniques can be understood and, with this, allows suggestions for modifications in the treatment protocols to the patient with lip and palatine cleft with an intention to minimize these negative effects on craniofacial growth.

References

- Lawson W, Patel ZM, Lin FY. The development and pathologic processes that influence maxillary sinus pneumatization. Anat Rec 2008;291(11):1554-63.
- 2. Alarcón KMG, Sá ÁJA. Epidemiological profile of patients

with orofacial cleft treated by a reference surgical team in the State of Amazonas, Brazil. Braz J Plast Sugery 2017;32(4):486-90.

- Erdur O, Ucar FI, Sekerci AE, Celikoglu M, Buyuk SK. Maxillary sinus volumes of patients with unilateral cleft lip and palate. Int J Pediatr Otorhinolaryngol 2015;79(10):1741-4. doi: http://dx.doi.org/10.1016/j.ijporl.2015.08.003
- 4. Ishikawa Y, Amitani R. Nasal and paranasal sinus disease in patients with congenital velopharyngeal insufficiency. Arch Otolaryngol Head Neck Surg 1994;120(8):861-5.
- Suzuki H, Yamaguchi T, Furukawa M. Maxillary sinus development and sinusitis in patients with cleft lip and palate. Auris Nasus Larynx 2000;27(3):253-6.
- Koppe T, Weigel C, Barenklau M, Kaduk W, Bayerlein T, Gedrange T. Maxillary sinus pneumatization of an adult skull with an untreated bilateral cleft palate. J Craniomaxillofac Surg 2006 Sep;34 Suppl 2:91-5.
- Demirtas O, Kalabalik F, Dane A, Aktan AM, Ciftci E, Tarim E. Does unilateral cleft lip and palate affect the maxillary sinus volume? Cleft Palate Craniofac J. 2018;55(2):168-72.
- Som P, Curtin H. Embryology, anatomy, physiology, and imaging of the sinonasal cavities. Head Neck Imaging 2014;99:166. doi: 10.1016/b978-0-323-05355-6.00002-1
- Robinson HE, Zerlin GK, Passy V. Maxillary sinus development in patients with cleft palates as compared to those with normal palates. Laryngoscope 1982;92(2):183-7. doi: 10.1002/lary.1982.92.2.183.
- Francis P, Raman R, Korula P, Korah I. Pneumatization of the paranasal sinusis (Maxillary and frontal) in cleft lip and palate. Arch Otolaryngol Head Neck Surg 1990;116(8):920-2. doi: 10.1001/archotol.1990.01870080042012.
- Kula K, Hale LN, Ghoneima A, Tholpady S, Starbuck JM. Cone-Beam Computed Tomography Analysis of Mucosal Thickening in Unilateral Cleft Lip and Palate Maxillary Sinuses. Cleft Palate Craniofac J 2016;53(6):640-8. doi: 10.1597/15-005
- Eckel W, Beisser D. Studies on the problem of the effect of harelip formation on the dimensions of the maxillary antra. Z Laryngol Rhinol Otol 1961;40:23-31.
- Ishikawa Y., Kawano M, Honjo I, Amitani R. The cause of nasal sinusitis in patients with cleft palate. Arch Otolaryngol Head Neck Surg 1989;115:442-6. doi: 10.1001/ archotol.1989.01860280040014.
- 14. T.D. Smith, M.I. Siegel, M.P. Mooney, A.M. Burrows J. Todhunter. Formation and enlargement of the paranasal sinuses in normal and cleft lip and palate human fetuses. Cleft Palate Craniofac J 1997;34:483-9. doi: 10.1597/1545-1569_1997_034_0483_faeotp_2.3.co_2.
- Suzuki H, Yamaguchi T, Furukawa M. Rhinologic computed tomographic evaluation in patients with cleft lip and palate. Arch Otolaryngol Head Neck Surg 1999;125:1000-4. doi: 10.1001/archotol.125.9.1000.
- Jaffe BF, Deblanc CB. Sinusitis in children with cleft lip and palate. Arch Otolaryngol 1971;93(5):479-82. doi: 10.1001/ archotol.1971.00770060725007
- 17. Harar R, Chadha N, Rogers G. The role of septal deviation in adult chronic rhinosinusitis: a study of 500 patients. Rhinology 2004;42:126-30.
- 18. Stallman J, Lobo J, Som P. The incidence of concha bullosa

and its relationship to nasal septal deviation and paranasal sinus disease. Am J Neuroradiol 2004;25:1613-8.

- 19. Boyce J, Eccles R. Do chronic changes in nasal airflow have any physiological or pathological effect on the nose and the paranasal sinuses? A systematic review. Clin Otolaryngol 2006;31:15-9. doi: 10.1111/j.1749-4486.2006.01125.x.
- Milczuk H, Dalley R, Wessbacher F, Richardson M. Nasal and paranasal sinus anomalies in children with chronic sinusitis. Laryngoscope 1993;103:247-52. doi: 10.1288/00005537-199303000-00002.
- 21. Salib R, Chaudri S, Rockley T. Sinusitis in the hypoplastic maxillary antrum: the crucial role of radiology in diagnosis and management. J Laryngol Otol 2001;115:676-8. doi: 10.1258/0022215011908612.
- 22. de Rezende Barbosa GL, Pimenta LA, Pretti H, Golden BA, Roberts J, Drake AF. Difference in maxillary sinus volumes of patients with cleft lip and palate. Int J Pediatr Otorhinolaryngol 2014;78(12):2234-6. doi: 10.1016/j. ijporl.2014.10.019.
- Hikosaka M, Nagasao T, Ogata H, Kaneko T KK. Evaluation of maxillary sinus volume in cleft alveolus patients using 3-dimensional computed tomography. J Craniofacial Surg 2013;24:23-6. doi: 10.1097/SCS.0b013e318267bdf3.
- 24. Norwak R V. X-ray Film Analysis of the Sinus Pnranasales from Cleft Patients (In Comparison with a Healthy Group). Anat Anz 1977;142:451-70.
- 25. Schneiderman ED, Xu H, Salyer KE. Characterization of the maxillary complex in unilateral cleft lip and palate using Cone-Beam computed tomography: a preliminary study. J Craniofac Surg 2009;20:1699-710. doi: 10.1097/ SCS.0b013e3181b3eddf.
- 26. Jakobsone G, Neimane L KG. Two- and three-dimensional evaluation of the upper airway after bimaxillary correction of Class III malocclusion. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;110:234-42. doi; 10.1016/j. tripleo.2010.03.026.
- 27. Lowe AA, Fleetham JA, Adachi S RC. Cephalometric and computed tomographic predictors of obstructive sleep apnea severity. Am J Orthod Dentofac Orthop 1995;107:589-95. doi: 10.1016/s0889-5406(95)70101-x
- 28. Lu Y, Liu Z, Zhang L, Zhou X, Zheng Q, Duan X, Zheng G WH, D H. Associations between maxillary sinus mucosal thickening and apical periodontitis using cone-beam computed tomography scanning: a retrospective study. J Endod 2012;38:1069-74. doi: 10.1016/j.joen.2012.04.027
- 29. Shanbhag S, Karnik P, Shirke PSV. Association between periapical lesions and maxillary sinus mucosal thickening: a retrospective cone-beam computed tomographic study. J Endod 2013;39:853–857. doi: 10.1016/j.joen.2013.04.010.
- 30. Vallo J, Suominen-Taipale L, Huumonen S, Soikkonen KNA. Prevalence of mucosal abnormalities of the maxillary sinus and their relationship to dental disease in panoramic radiography: results from the Health 2000 Health Examination Survey. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;109:80-7. doi: 10.1016/j.tripleo.2009.10.031.
- Rachelefsky GS, Katz RM SS. Chronic sinusitis in the allergic child. Pediatr Clin North Am 1988;35:1091-101. doi: 10.1016/s0031-3955(16)36550-6.
- 32. Wojciech D, Olivera O, Shetye P, Cutting C, Grayson B WS. Cleft palate midface is both hypoplastic and displaced. J Craniofac Surg 2013;24:89-93. doi: 10.1097/ SCS.0b013e3182646273.

- 33. Da Silva Filho OG, Calvano F, Alcoforado Assunção AG, Cavassan ADO. Craniofacial morphology in children with complete unilateral cleft lip and palate: a comparison of two surgical protocols. Angle Orthod 2001;71(4):274-84. doi: 10.1043/0003-3219(2001)071<0274:CMICWC>2.0.CO;2.
- 34. Capelozza Filho L, Normando AD SFO. Isolated influences of lip and palate surgery on facial growth: comparison of operated and unoperated male adults with UCLP. Cleft Palate Craniofac J 1996;33:51-6. doi: 10.1597/1545-1569_1996_033_0051_iiolap_2.3.co_2.
- Silva Filho OG, Lauris RCMC CA. Cleft lip and palate patients: effects caused by cheiloplasty. Ortodontia 1990;23.
- RB R. Treatment variables affecting facial growth in complete unilateral cleft lip and palate. Part 1. Treatment affecting growth. Cleft Palate J 1987;24:5-77.
- 37. Leonard AG, Kneafsey B, McKenna S, Johnston CD BD. A retrospective comparison of craniofacial form in

Northern Palate, with unilateral cleft lip and palate. Cleft Palate Craniofac J 1998;35:402-7. doi: 10.1597/1545-1569 1998 035 0402 arcocf 2.3.co 2.

- 38. Pradel W, Senf D, Mai R, Ludicke G, Eckelt U LG. Onestage palate repair improves speech outcome and early maxillary growth in patients with cleft lip and palate. J PhysiolPharmacol 2009;60(8):37-41.
- 39. Dissaux C, Grollemund B, Bodin F, Picard A, Vazquez MP MB et al. Evaluation of 5-year-old children with complete cleft lip and palate: multicenter study. Part 2: functional results. J Craniomaxillofac Surg 2016;44(2):94-103. doi: 10.1016/j.jcms.2015.08.029
- 40. Schilling GR, Cardoso MC de AF, Maahs MAP. Effect of palatoplasty on speech, dental occlusion issues and upper dental arch in children and adolescents with cleft palate: an integrative literature review. Rev CEFAC 2019;21(6). doi: 10.1590/1982-0216/201921612418