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Multidisciplinary Treatment in a Patient with Bilateral Cleft Lip and Palate: A Case Report

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Abstract

Aim: Cleft lip and palate (CLP) are among children's most common congenital craniofacial abnormalities. CLP results from the failure of the fusion of facial processes during embryogenesis. The etiology of CLP is complex and thought to involve genetic influences along with variable interactions from environmental factors. The treatment process involves a multidisciplinary approach while focusing on achieving functional and aesthetic well-being for the patient.
Case Report: In this paper, we report using a multidisciplinary approach to treat a complex case of bilateral CLP in a 7-year-old female patient. The treatment significantly changed the patient's facial profile, in addition to improving her occlusion, aesthetics, and function.
Conclusion: The treatment of CLP should follow a multidisciplinary approach while considering several factors, such as age, developmental stage, and psychosocial factors. This approach has been shown to improve patients' well-being and quality of life.

Keywords: Orthodontic Treatment, Cleft Palate, Malocclusion.

Background

Clefts of the lip and palate (CLP) are among children's most common congenital craniofacial anomalies (1). The worldwide incidence of cleft lip with or without cleft palate is 1 in 700 births. This condition is more frequent in males than females (2,3). CLP results from the failure of fusion of the medial nasal process with the maxillary process, which occurs between the 5th and 10th weeks of intrauterine development (2,4). The etiology of CLP is complex, involving genetic influences with variable interactions from environmental factors (5).

Treatment of CLP begins with surgical repair during the first few months of life to correct or reconstruct the palate (6). This correction might cause scar tissues, limiting the growth of the impacted area and leading to decreased transverse and anteroposterior dimensions of the maxilla (6). As a result, patients with CLP often have transverse maxillary atresia and might develop maxillary retrusion, anterior crossbite, unilateral or bilateral posterior crossbite, and compromised airway development, among other dentoskeletal discrepancies (6). These changes can significantly impact the psychological development and quality

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of life of patients with CLP (7). During the full-mouth fixed orthodontic treatment stage, it is usually combined with orthognathic surgery, correcting severe jaw discrepancy (3).

Orthodontic treatment is vital in the multidisciplinary therapeutic approach for patients with CLP (8). Generally, orthodontic intervention is carried out in childhood and adolescence and requires good commitment and cooperation from the patient and family members, as the treatment can be lengthy and complex, depending on the severity of the malformation and its impact on the occlusion and masticatory and chewing functions (8).

Dental alterations are more frequent in patients with CLP compared to the general population (9). The most common alterations include agenesis of the upper lateral incisors, supernumerary teeth, rotation, and changes in tooth shapes (10). These patients may require interventions from various dental specialties to achieve satisfactory outcomes (11).

In addition to anatomical and functional alterations, CLP might impact facial aesthetics and normal speech development, resulting in potentially lifelong social and psychological consequences (12). Given the broad impact of CLP on the human body, the treatment of CLP should apply a multidisciplinary approach across various specialties to address the patient's physical well-being and contribute to a successful CLP treatment outcome (11,13)

Case Report

A 7-year-old female patient with bilateral CLP was referred by the clinical dentist in 2005 to undergo orthodontic treatment and subsequent bone graft surgery.

The patient's medical history did not reveal any relevant health condition. An extraoral evaluation revealed hypotonia of the orbicularis muscles, mouth breathing, atypical swallowing and wheezing, a symmetric face, proportional facial thirds, a concave facial profile, a retruded upper lip, and a protruded lower lip.

Intraoral analysis showed that the patient was in the mixed dentition phase and presented an Angle Class I malocclusion, combined with a Class II canine relationship, a transverse maxillary deficiency, a mandibular arch with a parabolic shape, an accentuated overbite, and an anterior crossbite (Figure 1).

A panoramic x-ray confirmed mixed dentition and the presence of all permanent teeth, except for teeth #12 and #22, with a diagnosis of agenesis of those elements (Figure 2). The cephalometric evaluation revealed a Class II skeletal malocclusion (ANB=5.2°), with maxillary and mandibular retrusion (SNA=80.8°, SNB=75.6°). The U1 to NA measurement of -5.9° indicated lingualized maxillary incisors, while the L1 to NB value of 20.4° indicated mandibular incisors with buccolingual inclination within the normal range. U1-NA value of -3.1 mm and a L1-NB value of 3.6 mm indicated retruded maxillary incisors and the normal position of the mandibular incisors, respectively. An SN-GoGn of 39.5° indicated a vertical growth pattern (Figure 3).







Figure 1. Pretreatment photographs



Figure 2. Pretreatment OPG

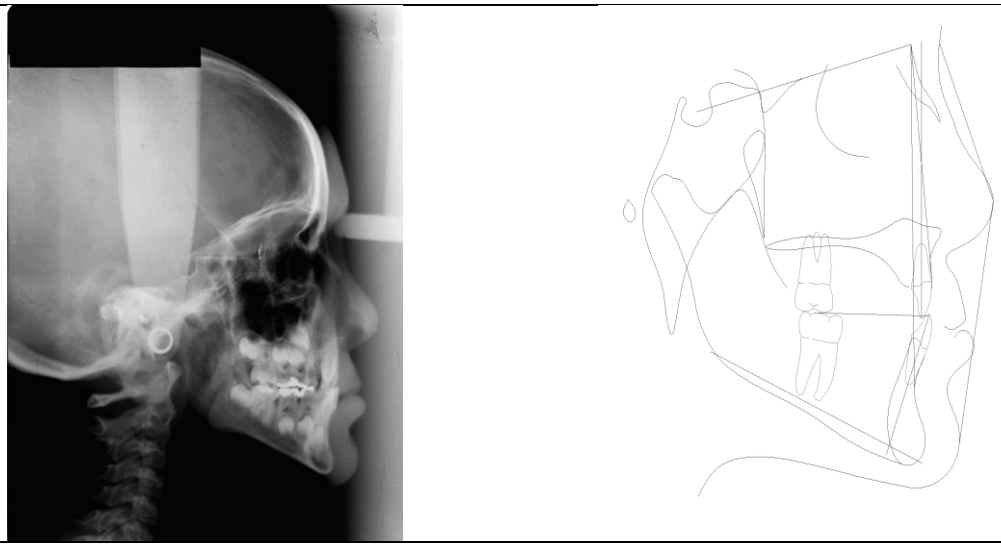


Figure 3. Patient's Initial lateral cephalogram and tracing

Treatment Objectives and Alternatives

The proposed treatment included a maxillary disjunction with a Haas expander appliance associated with a maxillary protraction headgear, the initial installation of a 2-by-2 fixed orthodontic appliance (with orthodontic accessories on the upper molars and upper central incisors), and bone grafting in the areas of the cleft.

The treatment began in August 2005 with the Haas palatine expander appliance to improve the maxillary atresia. A posterior bite stop was used to lift the bite and allow the maxilla to advance, overlapping the anterior bite. After two months, the partial bonding of the fixed appliance began, and a wire with a projection and expansion spring was installed to project teeth #11 and #21.

In March 2006, a quad helix appliance was used in association with a headgear for maxillary protraction (Figures 4 to 6). The headgear was suspended during the following year after 3 mm of horizontal overlap was achieved. A transpalatal bar was then installed to preserve the transverse dimension. Bone grafting was performed on the left side to support the teeth in the cleft area and allow tooth movement and traction of tooth #22.

In April 2009, another disjunction of the maxilla was performed, in addition to the nocturnal use of elastics on teeth #16 and #46 to address the overlapping teeth (Figure 7).



Figure 4. Intermediary cephalometric x-ray profile taken in 2006.



Figure 5. Periapical x-rays taken in 2006.



Figure 6. Intermediary panoramic x-ray taken in 2006.



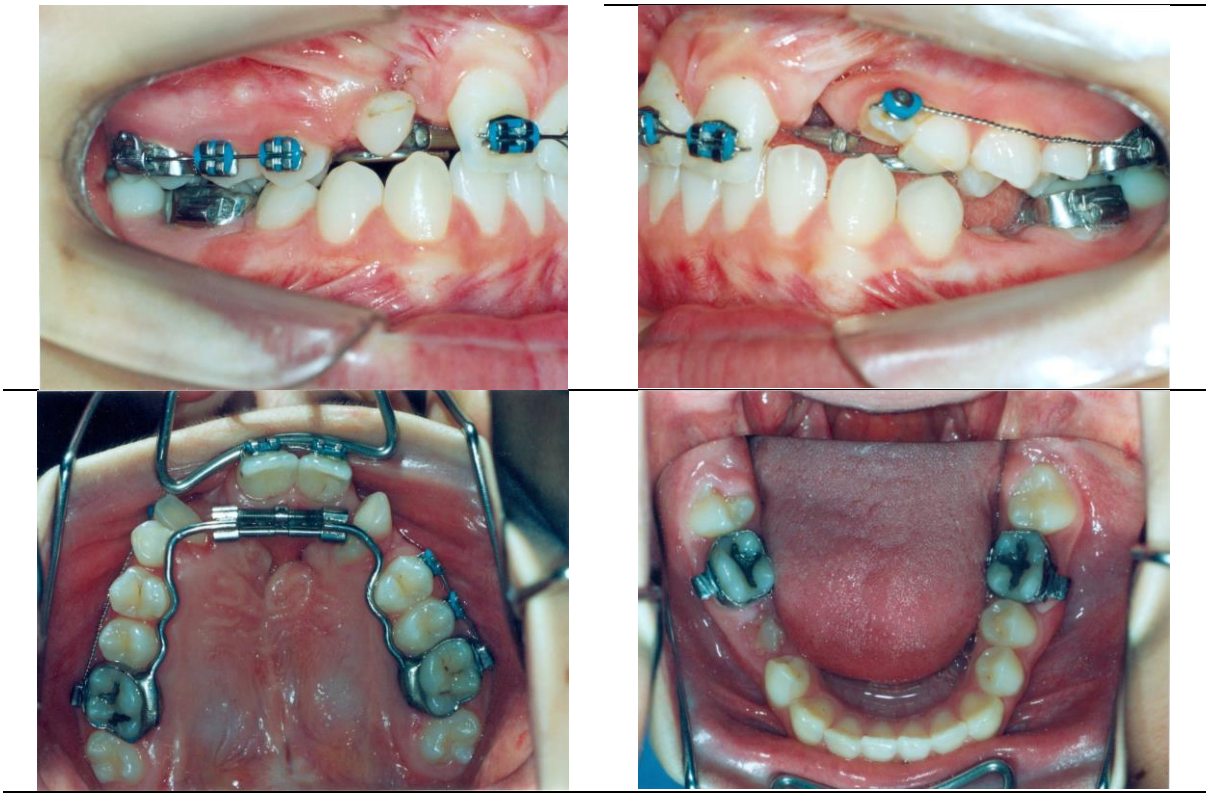


Figure 7. Intermediary intraoral photographs taken in 2009.

In the following year, the patient underwent a speech-language evaluation where the orofacial structures (tongue, palate, palatine tonsils, and pharynx) were assessed. The diagnosis was velopharyngeal incompetence due to increased intraoral pressure. Exercises were prescribed to decrease intraoral pressure, create an intraoral negative pressure, and improve palate strength and swallowing.

The patient continued treatment with a fixed appliance for another two years when tooth #63 was extracted. A new evaluation with a speech therapist detected a decrease in hypernasality, suggesting continuity of treatment along with exercises to increase intraoral pressure. In the following year, new periapical x-rays were ordered, which showed insufficient bone mesial to tooth #13. In 2013, bilateral graft surgery was performed to correct the bone defects caused by the bilateral cleft.

After the interceptive orthodontic treatment combined with bone graft surgeries, the corrective phase began for aligning and leveling the teeth, correcting tooth positions, and distributing spaces to allow the future placement of implants in the region of teeth #12 and #22. A temporary tooth was added to the fixed appliance to provide an adequate aesthetic effect (Figure 8). Once the patient's craniofacial growth was completed, dental implants were placed in the region of teeth #12 and #22 using a provisional prosthesis. Subsequently, definitive prostheses were placed on implants, along with cosmetic reconstructions with composite resin in teeth #13 and #23 (Figure 9).



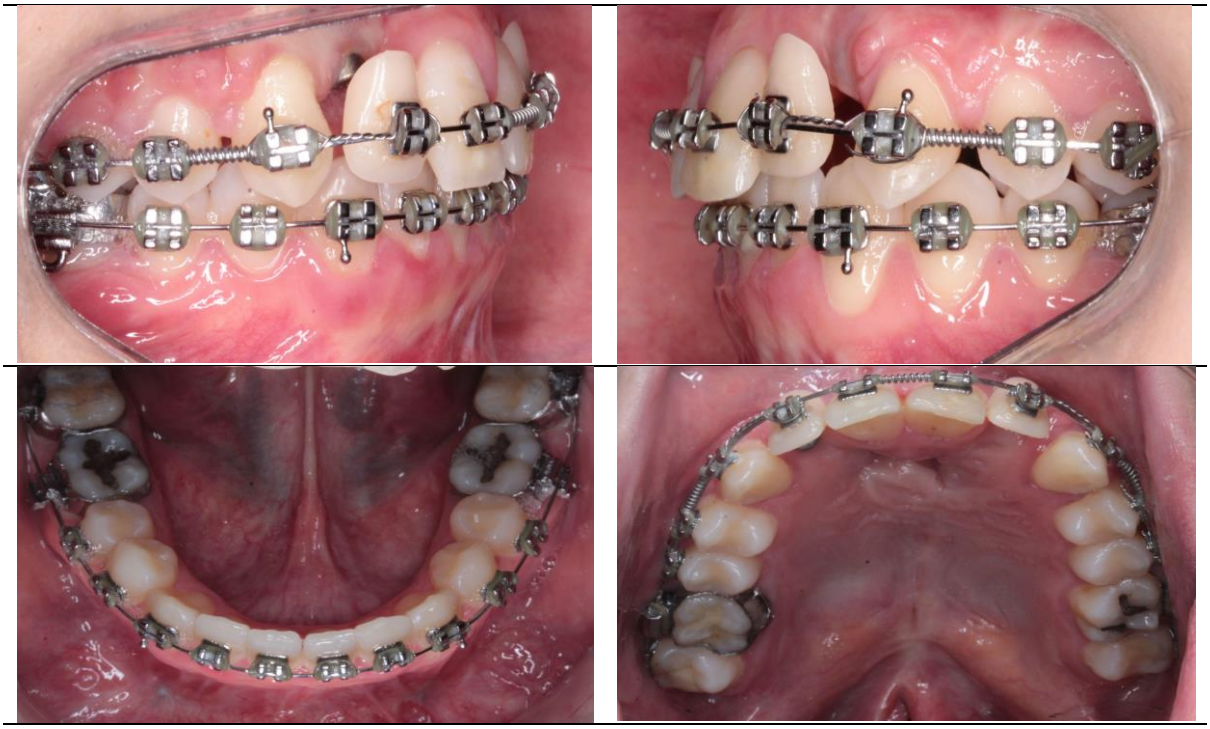


Figure 8. Intermediary intraoral photographs taken in 2021.

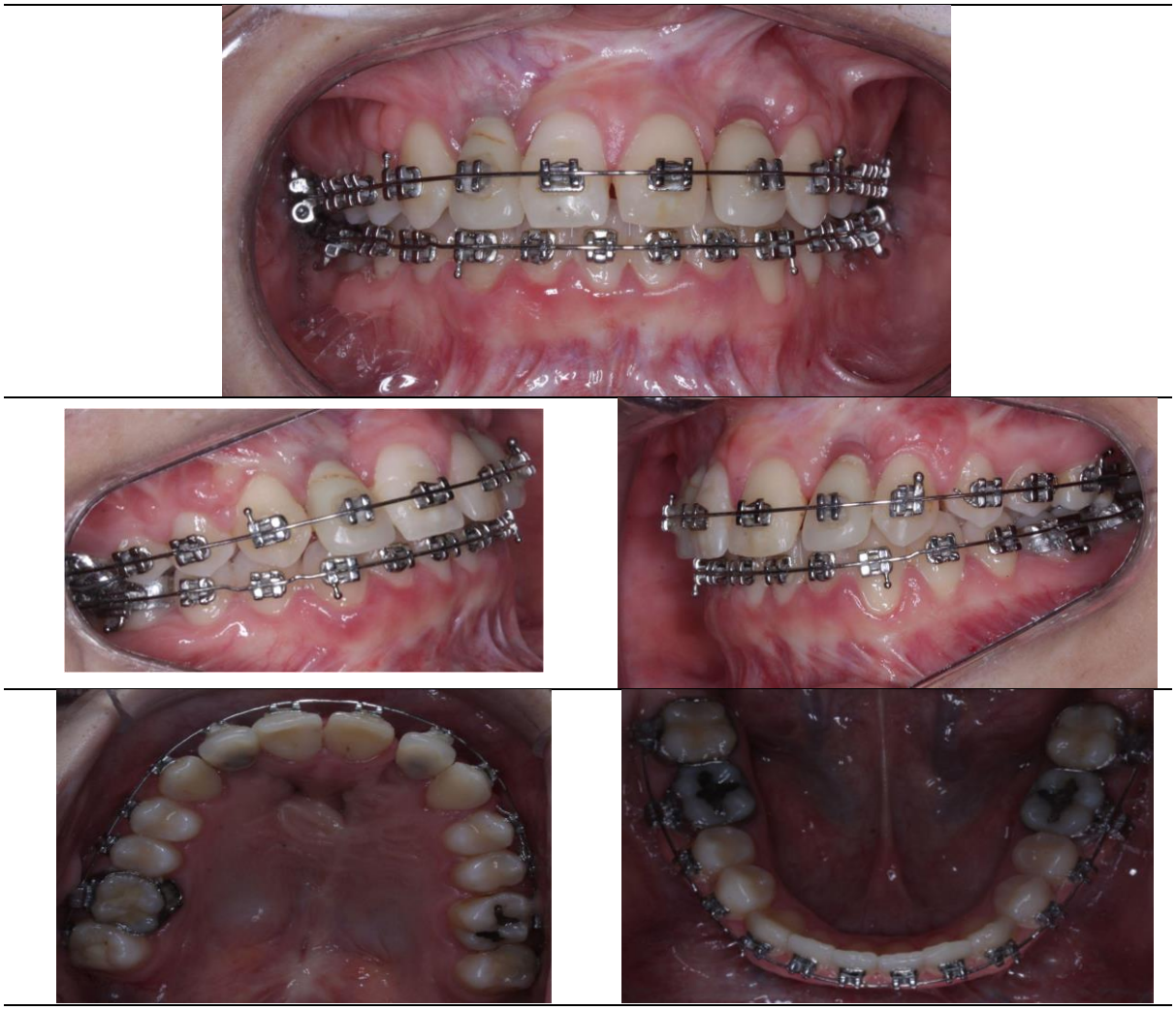


Figure 9. Intermediary intraoral photographs taken in 2022.

Treatment Results

The treatment was completed in September 2022; the facial symmetry and proportionality of the facial thirds were maintained. In the intraoral evaluation, the patient presented parabolic upper and lower arches. The transverse gain obtained with the treatment was observed in the frontal view. In the lateral view, crossbite correction was noted, with horizontal and vertical overbites within normal limits (Figure 10).



Figure 10. Final facial and intraoral photographs.

These findings demonstrated the benefits of the orthodontic treatment, including normal maxillary transverse dimension, improvement in the molar and canine relationships, satisfactory overbite and overjet, changes in the shape of the arches, which are now parabolic, and good tooth engagement. In addition, an improvement in the alignment of the incisors in their bone bases was observed, contributing to a favorable occlusion. A straight facial profile and facial symmetry were observed, along with a normal smile, harmony of the buccal corridor, and improved nasolabial angle due to the advancement of the maxilla.

The patient also achieved considerable

functional improvement, including better sibilant phonation and improved swallowing, which were previously abnormal. Improved self-esteem and overall satisfaction with the treatment were also reported, indicating a positive impact on the patient's psychosocial aspect.

The final panoramic x-ray evaluation showed no signs of root resorption, adequate overall alveolar bone levels, and good root parallelism (Figure 11). Post-treatment cephalometric evaluation (Figure 12) showed an SNA of 77.1° and SNB of 76.7°, indicating the preservation of the maxillary and mandibular retrusion characteristics. The ANB value of 0.4° demonstrated improvement from a Class II to Class I skeletal profile. The 1.NA measurement of 36.6° indicated lingualized upper incisors, whereas the 1.NB measurement of 20.4° indicated lower incisors with buccolingual inclination within the normal range. The 1-NA value of 9.0 mm and 1-NB value of 4.3 mm indicated protruded upper incisors and well-positioned lower incisors, indicating an improvement in their positions. The SN-Go.Gn value of 33.7° indicated a normal growth pattern.



Figure 11. Final panoramic x-ray after treatment.

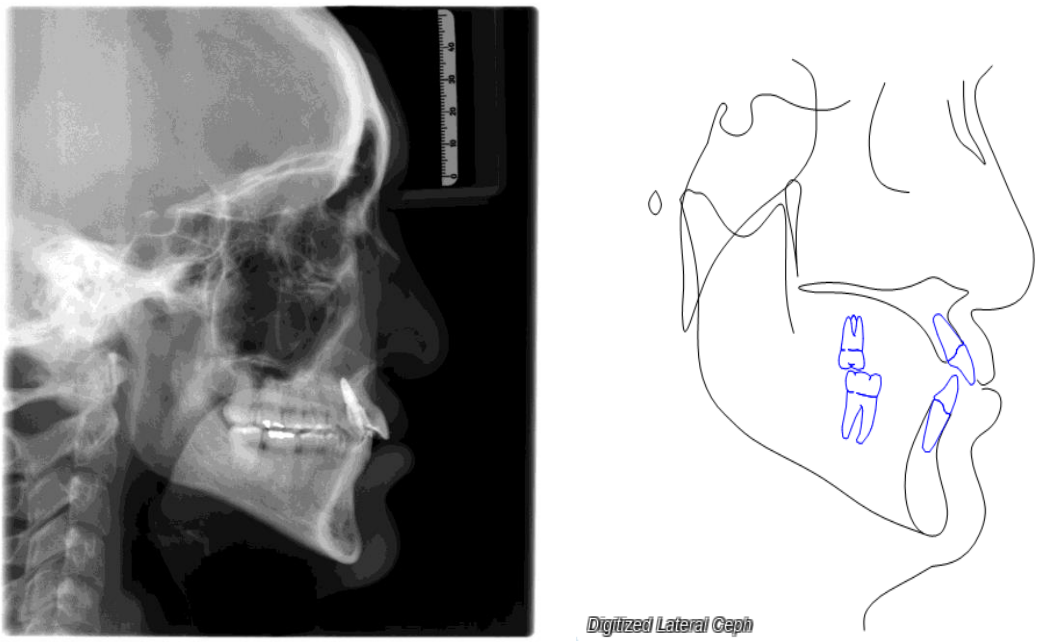


Figure 12. Final cephalometric x-ray profile and cephalometric tracing.

Discussion

Patients with CLP are more susceptible to skeletal problems compared to non-cleft patients (14). These individuals may frequently present with midface deficiencies, dental crossbites, dental anomalies, intraoral and extraoral soft tissue asymmetries, poor soft tissue healing, and problems with eating and speaking. In this reported case, the patient had bilateral CLP and presented several clinical conditions consistent with the literature, such as deficiency in maxillary growth, a concave profile, dental agenesis, and wheezing.

Due to the alterations presented and the high complexity of cases involving CLP, the treatment of this condition is usually long (14) and often performed at different stages of development, which vary according to cleft severity, amount of skeletal discrepancies, and the presence of dental abnormalities (13).

The therapeutic intervention for individuals with CLP begins during the early months of infancy through surgical procedures aimed at repairing the clefts in the lip and palate, thereby facilitating the consolidation of the cleft segments. This union not only diminishes the transverse dimension of the maxilla but also engenders fibrous tissue formation, with a constraining influence on the typical developmental processes within the affected anatomical region (6).

In addition to surgical and orthodontic treatments (15), other therapeutic approaches are often applied. Orthodontics is considered a vital component of the non-invasive treatment that meets the patient's primary needs, improving quality of life and preparing the individual for other procedures that may be indicated throughout the treatment (15).

Orthodontic intervention is usually initiated in the mixed dentition phase, as during this period, the orthopedic response is more favorable, avoiding relapses with growth (13). In the present case, the patient was initially submitted to an interceptive treatment and, subsequently, to a corrective treatment. The overall treatment took approximately 20 years to complete. During this period, in addition to the orthodontic treatment, bone grafting was performed in the cleft areas to allow for tooth movement and restore bone integrity. Implants were installed to correct the presented agenesis impacting teeth #12 and #22, as well as restorative treatments.

A multidisciplinary therapeutic approach involving the support of psychologists, orthodontists, surgeons, geneticists, pediatricians, ear, nose, and throat specialists, anesthetists, otorhinolaryngologists, nurses, nutritionists, speech therapists, among others, may be necessary to meet all the demands of patients with CLP (16). In the present report, the CLP patient received care from several healthcare specialties throughout the treatment, including the speech-language pathology team, which played a significant role in diagnosing velopharyngeal insufficiency. This condition is characterized by speech alterations caused by the congenital anomaly itself and by the surgeries performed to repair the clefts, which led to reduced dimensions of the nasal cavity and decreased function of the lower airways (17). Due to the combined orthodontic and speech therapy approaches, continuation of the treatment of velopharyngeal incompetence was suggested, which resulted in improved hypernasality and overall improvements in speech and respiratory functions.

The approaches used to treat the patient's CLP resulted in increased maxillary transverse dimension and improved facial features and tooth alignment. A multidisciplinary therapeutic approach promoted adequate occlusion, function, and aesthetics. This approach should consider that patients may have different needs and require long-term care to improve well-being and quality of life at all stages of treatment (18).

Conclusion

A multidisciplinary approach should be followed to manage CLP. This approach should consider several factors, including the patient's condition and needs, age, stage of development, and psychosocial factors. Management of complex cases often requires lengthy treatments to continuously improve the patient's well-being throughout the different developmental stages. In the present case report, successful results were obtained after a multidisciplinary approach was applied to manage the patient's CLP. This included achieving satisfactory aesthetics, functional, and occlusion results and ensuring minimal impact on the patient's psychological well-being throughout the treatment.

Conflicts of interest

The authors have no relevant financial or non-financial interests to disclose

Data Availability

The data supporting the findings of this study are available from the corresponding author upon reasonable request.

Acknowledgments
Not applicable

References

1. Neville BW, Damm DD, Allen CM, Chi AC. Oral and maxillofacial pathology. 4th edition. Philadelphia: Elsevier Inc. 2016; P: 1-3.
2. Costa VCR, da Silva RC, de Oliveira IF, Paz LB, Pogue R, Gazzoni L. [Etiological aspects and clinical trials of Labiopalatine]. Rev Med Saude Brasilia. 2018;7(2):258-68.
3. Chen YH, Liao YF, Chang CS, Lu TC, Chen KT. Patient satisfaction and quality of life after orthodontic treatment for cleft lip and palate deformity. Clin Oral Invest. 2021;25(9):5521-9. doi:10.1007/s00784-021-03861-4.
4. Francisco I, Caramelo F, Fernandes MH, Vale F. A Comparative Study of Oral Health-Related Quality of Life among Cleft Lip and Palate Patients and Their Families during Orthodontic Treatment. Int J Environ Res Public Health. 2021;18(23):12826. doi:10.3390/ijerph182312826.
5. Figueiredo MC, Pinto NF, Fabricio, FK, Boaz, CMS, Faustino-Silva DD. [Patients with cleft lip and palate – clinical cases follow up] Conscientiae Saúde. 2010; 9(2): 300-8. doi:10.5585/conssaude.v9i2.2256.
6. de Sousa AA, Cheib PL, Júnior IA, Oliveira DD, Souki BQ, Cevidanes LHS. [Comparison of two expanders in maxillary expansion in cleft patients: report of two cases]. Rev Port Estomatol Med Den Cir Maxilofac.2016;57(2):116-24. doi:10.1016/j.rpemd.2016.01.003.
7. Cassi D, Di Blasio A, Gandolfinini M, Magnifico M, Pellegrino F, Piancino MG. Dentoalveolar Effects of Early Orthodontic Treatment in Patients With Cleft Lip and Palate. J Craniofac Surg. 2017;28(8):2021-6. doi:10.1097/SCS.0000000000003854.
8. Crerand CE, Da Silveira AC, Kapa H, et al. Adherence to Orthodontic Treatment in Youth With Cleft Lip and/or Palate. Cleft Palate Craniofac J. 2020;57(2):218-27. doi:10.1177/1055665619867556.
9. Ezzeldin M, Gee S, Curtis J, Clark VJ, Smallridge J, Collard M. Dental anomalies in cleft lip and/or palate children at age 10 - a retrospective review across three cleft centres: Part 1. Br Dent J. 2023;234(12):926-30. doi:10.1038/s41415-023-5976-5.
10. Sá J, Araújo L, Guimarães L, Maranhão S, Lopes G, Medrado A, Coletta R, Reis S. Dental anomalies inside the cleft region in individuals with nonsyndromic cleft lip with or without cleft palate. Med Oral Patol Oral Cir Bucal. 2016 ;21(1):e48-52. doi: 10.4317/medoral.20757.
11. Cumerlato CBDF, Santos CSD, Santos MBFD, Bergoli CD, Boscato N. Multidisciplinary Approach for Reestablishing Function and Aesthetic of Unilateral Cleft Lip Defect: A Case Report. Cleft Palate Craniofac J. 2021;58(3):396-9. doi:10.1177/1055665620946560.
12. Prado JP, Reus JAB, Soares AF, Lemos GCSM. [Demystifying Labiopalatine Fissure]. Rev Mult Psic. 2018. 12(42):229-41. doi:10.14295/idonline.v12i42.1433.
13. Parsaei Y, Uribe F, Steinbacher D. Orthodontics for Unilateral and Bilateral Cleft Deformities. Oral Maxillofac Surg Clin North Am. 2020;32(2):297-307. doi:10.1016/j.coms.2020.01.011.
14. Hameed O, Amin N, Haria P, Patel B, Hay N. Orthodontic burden of care for patients with a cleft lip and/or palate. J Orthod. 2019;46(1):63-7. doi:10.1177/1465312518823010.
15. Agrawal AB, Kidiyoor H, Patil Morth AK. Treatment of Unilateral Cleft Lip and Palate Patient with Intraoral Tooth Tissue Borne Distractor and Facemask Therapy: A Case Report. Cleft Palate Craniofac J. 2021;58(12):1569-76. doi:10.1177/1055665621993366.
16. Hodgkinson PD, Brown S, Duncan D, Grant C, Mc Naughton A, Thomas P, Mattick CR. Management of children with cleft lip and palate: a review describing the application of multidisciplinary team working in this condition based upon the experiences of a regional cleft lip and palate centre in the United Kingdom. Fetal Matern Med Rev. 2005;16(1):1-27. doi:10.1017/S0965539505001452.
17. Alrejaye N, Gao J, Hatcher D, Oberoi S. Effect of maxillary expansion and protraction on the oropharyngeal airway in individuals with non-syndromic cleft palate with or without cleft lip. PLoS One. 2019;14(7):e0213328. doi:10.1371/journal.pone.0213328.
18. Padovano WM, Snyder-Warwick AK, Skolnick GB, et al. Evaluation of Multidisciplinary Team Clinic for Patients with Isolated Cleft Lip. Cleft Palate Craniofac J. 2020;57(7):900-8. doi:10.1177/1055665619900625.