CASE REPORT

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Orthodontic Treatment of Unilateral Cleft Lip and Palate Associated with Agenesis of Maxillary Lateral Incisors: A Case Report

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ABSTRACT

Cleft lip and palate (CLP) is a common congenital condition presenting significant orthodontic challenges due to maxillofacial growth disturbances, associated malocclusions, and dental anomalies. Patients with CLP often have complex dental and skeletal issues, requiring a comprehensive, interdisciplinary approach to address functional and aesthetic concerns. An 11-year-old female presented with unilateral cleft lip, alveolus, and palate (UCLP) associated with dental anomalies, which included a skeletal Class I relationship with Angle Class II malocclusion, bimaxillary retrognathia, bidental retroclination, negative overjet, lower dental midline shifting, anterior crossbite, microdontia #12, agenesis #22, and several malpositioned teeth. The patient was treated with fixed orthodontic appliances, including the extraction of remaining deciduous teeth, mesialisation of the teeth on the upper left side to close the space resulting from agenesis, and space management in the mandible. Significant improvements were observed in overjet, dental alignment, occlusion, and aesthetics after a 17-month treatment period. Following orthodontic treatment, restorative procedures were performed on teeth #12 and #23. Adhering to the prescribed retainer schedule is essential to preserving dental esthetics and the stability of the treatment results. Comprehensive orthodontic treatment with fixed appliances effectively addressed the complex dental and skeletal issues of this patient with UCLP. The treatment led to improved dental function and aesthetics, highlighting the importance of a personalised approach in addressing cleft cases.

Keywords: *Cleft lip and palate; comprehensive orthodontic treatment; dental agenesis; orthodontics; treatment outcome*

INTRODUCTION

Cleft lip and palate (CLP) represent classifications of craniofacial congenital anomalies that impact numerous children worldwide each year. In 2002, the World Health Organization published a report that cleft lip with or without accompanying cleft palate represents a significant congenital anomaly that impacts about 1 in every 600 neonates globally. CLP is a worldwide concern, with a child being born with some type of orofacial cleft nearly every three minutes. Over 10 million individuals globally are impacted by the condition (Shaw, 2004; Sandy *et al.*, 2020). Over a five-year period, nationwide research in Indonesia revealed a 0.04% increase in the prevalence of orofacial clefts. The national prevalence of cleft lip in Indonesia is 0.2%, as stated in the National Guidelines for Medical Services for the treatment of cleft lip and palate. Indonesia reports 7,500 cases of orofacial clefts annually, signifying a considerable prevalence that necessitates ongoing research in this area (Putri *et al.*, 2024). CLP occurs in 50% of people, typically due to the inadequate fusion of facial features prior to palate development. The reasons are classified into non-genetic variables, including environmental impacts (e.g., smoking, alcohol intake), and genetic factors, which encompass clefts linked to other malformations or occurring as isolated cases (Zaaba *et al.*, 2023; Kulesa-Mrowiecka *et al.*, 2024).

Orofacial clefts are a heterogeneous group of disorders affecting the structure of the face and oral cavity. They are divided into three general categories: clefts affecting the lip only (CL), clefts affecting the lip and palate (CLP), and clefts affecting the palate alone (CP). The condition may present as unilateral, bilateral, or midline and can be complete, incomplete, or submucous. Clefts can involve the nasal tip, philtrum, lip, lip vermillion, alveolus (gum), hard palate, soft palate, or uvula (Leslie & Marazita, 2013; Sundoro *et al.*, 2024). Due to the complex nature of the condition, which affects various aspects of a child's health and development, since the early 1950s, there has been a prevailing agreement that children who are affected by cleft conditions require thorough and coordinated management by a team of interdisciplinary professionals. The team offers comprehensive cleft care and typically incorporates specialists from the following disciplines: paediatrics and obstetrics, plastic and reconstructive surgery, orthodontics, genetics, social work and/or nursing, ear nose and throat, speech/language pathology, maxillofacial surgery, prosthetic dentistry, and psychology (Kuijpers-Jagtman, 2006; Kuijpers-Jagtman & Kuijpers, 2023).

Orthodontic treatment for patients with CLP is a multifaceted process that requires careful planning and collaboration to address the dental, skeletal, and functional discrepancies associated with CLP, ultimately improving the patient's aesthetics, function, and quality of life (Iswati *et al.*, 2023). Early orthodontic intervention plays a crucial role in the management of CLP. The orthodontic intervention for patients with CLP during deciduous and mixed dentition stages has been advocated to establish more advantageous conditions for midfacial development, normalise the intermaxillary basal relationship, and eliminate functional anomalies (Cassi *et al.*, 2017; Gopinath *et al.*, 2017).

The most severe type of cleft is the complete cleft of the lip, alveolus, and palate, which can be either unilateral CLP (UCLP) or bilateral CLP (BCLP). In patients with UCLP, dental issues may refer to changes in the number of teeth, shape, and the time of the eruption. Potential factors contributing to tooth agenesis within or beyond the cleft region include abnormalities during embryonic development and/or possible iatrogenic complications arising from surgical procedures in the cleft area. Surgical procedures during the early stages of tooth development contribute to tooth agenesis in the cleft region, whereas agenesis in areas outside the cleft is predominantly associated with genetic factors or gene regulation. In addition to their significance in tooth

formation, these variables are also crucial to palatogenesis (Bartzela *et al.*, 2013). Agenesis of the maxillary lateral incisor in the cleft area is frequently observed in individuals with UCLP. After secondary alveolar bone graft surgery, the gold standard treatment plan is the mesial movement of maxillary canines to replace the absent lateral incisor (Manfio *et al.*, 2023). Another prevalent dental issue is ectopic eruption, which refers to the canine at the cleft side erupting at the palatal side (Paradowska-Stolarz *et al.*, 2022). This case report highlights the importance of a comprehensive approach for effectively treating a UCLP patient with agenesis of the cleft-sided maxillary lateral incisor to improve the patient's overall functional, structural, and aesthetic outcomes.

CASE REPORT

This work was reported in accordance with the CARE guidelines (Gagnier *et al.*, 2014). Written informed consent was obtained from the patient for publication of this case report and the accompanying images. An Indonesian 11-year-old female came to the dental hospital for orthodontic purposes. Her chief complaint was crowded front upper jaw teeth, and she was concerned about her facial appearance. She was born with a right-sided unilateral cleft lip, alveolus, and palate (UCLP). Primary labioplasty was performed when she was four months old by a plastic surgeon. Then, at the age of 17 months, she underwent a palatoplasty procedure with a plastic surgeon. Currently, the palatal cleft has been completely closed. No alveolar bone grafting was performed. No fistula is observed on either the buccal vestibule or at the palatal side. The patient denies any history of ear infections, there are no speech disturbances, and the patient's voice does not sound nasal.

She had a brachycephalic head type and a mesoprosope facial type. She is retrognathic in the midface. From frontal appearance, the patient showed scarring of the right upper lip and an asymmetry of the nose (Figs. 1a, 1b). Functional analysis revealed that the freeway space was normal (2.5 mm) with no evidence of temporomandibular disorder. Intraoral examination revealed a bilateral Angle Class II relationship with a negative overjet up to –1.1 mm, microdontia #12, agenesis #22, upper dental midline shifted to the right by 2 mm, anterior crossbite #11 to #42 and #13 to #43, and several malpositioned teeth (Fig. 2a). Moderate crowding with multiple diastema due to microdontia #12, and agenesis #22 resulting in a maxillary arch length discrepancy of +13.75 mm, whereas the mandibular arch showed slight anterior crowding (arch length discrepancy + 5.07 mm). Postoperative scarring of the palate is mild (Figs. 3a, 3b). So even though the upper jaw has a considerable arch length discrepancy, the transverse dimension has a good prognosis.

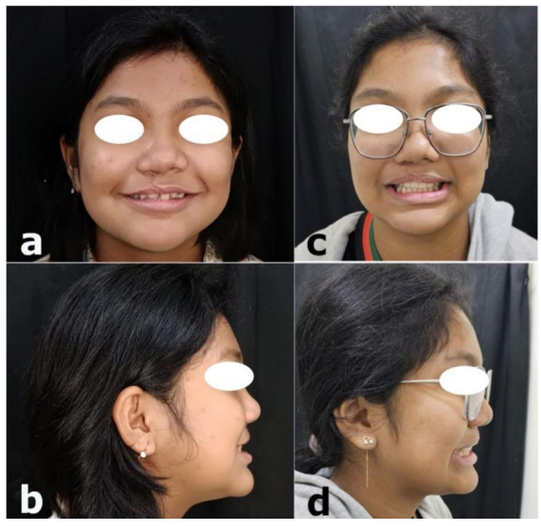

Fig. 1 (a) Extraoral photographs: pre-treatment (frontal); (b) Extraoral photographs: pre-treatment (lateral); (c) Extraoral photographs: post-treatment (frontal); and (d) Extraoral photographs: post-treatment (lateral).

Fig. 2 (a) Intraoral photographs: pre-treatment; (b) Intraoral photographs: during treatment stage; and (c) Intraoral photographs: after orthodontic treatment and conservative rehabilitation.

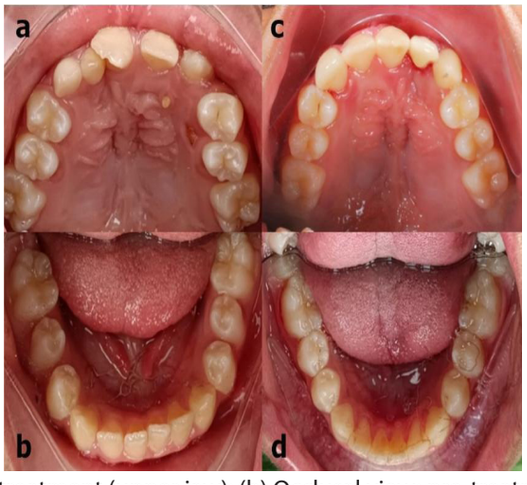

Fig. 3 (a) Occlusal view: pre-treatment (upper jaw); (b) Occlusal view: pre-treatment (lower jaw); (c) Occlusal view: after orthodontic treatment and conservative rehabilitation (upper jaw); and (d) Occlusal view: after orthodontic treatment and conservative rehabilitation (lower jaw).

The cephalometric findings revealed a skeletal Class I relationship (ANB 2.28°; Wits appraisal 2.13°) with a normal vertical facial growth pattern (SN-MP 32.54°) even though both maxilla and mandible are positioned retrognathic (SNA 76.25°; SNB 73.96°). The maxillary incisors are extremely retroclined (U1-palatal plane 101.6°; U1-NA 1.28 mm), while the mandibular incisors are still within the normal range (L1-MP 90.82°; L1-NB 4.05 mm). Rickett's lip analysis indicated a retrusive upper and lower lip (Table 1; Fig. 4a). The panoramic radiograph indicated an absence of the maxillary left lateral incisor (#22) and persistence of the upper right deciduous canine (#53). The alveolar cleft between teeth #11 and #13 is still present (Fig. 5a), and tooth #12 is located distally to the cleft.

Table 1 Lateral cephalometric measurements

Parameters	Normal (mean+SD)	Pre-treatment	Post-treatment
Horizontal skeletal			
SNA (°)	82±2	73.85	75.39
SNB (°)	80±2	73.06	73.75
ANB (°)	2±2	2.28	1.65
Wits appraisal (mm)	1±1	2.13	(–) 0.87
Angle of convexity (°)	0±5	2.02	0.95
Vertical skeletal			
Y-axis (°)	60±4	62.60	63.17
SN-mandibular plane (°)	32±3	32.54	32.99
FMPA (°)	27±5	26.15	25.30
Dental			
Interincisal angle (°)	135±10	142.37	132.41
U1-palatal plane (°)	109±6	101.60	104.35
U1-NA (mm)	4±2	1.28	4.84
L1-mandibular plane (°)	90±4	90.82	94.88
L1-NB (mm)	4±2	4.05	5.01
Soft tissue			
Upper lip to E-Line (mm)	1±2	(–) 2.94	(–) 4.01
Lower lip to E-Line (mm)	0±2	(–) 2.04	(–) 1.57

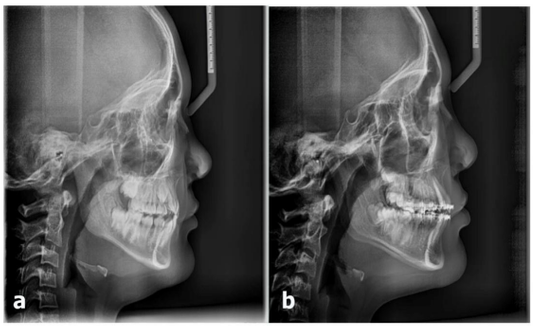

Fig. 4 (a) Cephalogram lateral: pre-treatment; and (b) Cephalogram lateral: post-treatment.

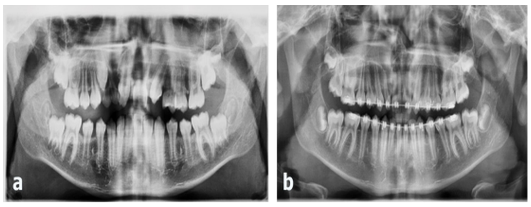

Fig. 5 (a) Panoramic pre-treatment; and (b) Panoramic post-treatment.

CASE MANAGEMENT

The initiation of the therapeutic intervention is dependent on the improvement of her oral hygiene. The steps that were carried out at the start and during the orthodontic treatment were educating, instructing, and motivating the patient, and explaining orthodontic care. An informed consent letter was signed after the patient's parents understood the treatment plan and agreed to all orthodontic treatment procedures. The treatment's objectives were to harmonise the facial profile by correcting the anterior crossbite, levelling and aligning the dental arches, and establishing a good interdigitation with enhanced intercuspation. The condition of the patient's alveolar cleft site after surgery was accepted by the medical team (the operator and supervising doctor), with no further intervention or surgery (such as alveolar bone grafting) deemed necessary. A treatment plan was suggested: (1) extraction of remaining deciduous tooth

(#53) and removal the radices of deciduous teeth; (2) alignment of the upper and lower teeth with Roth technique straightwire fixed appliance; (3) occlusal adjustment; and (4) retention using upper and lower Hawley retainer; (5) followed by restorative/prosthetic rehabilitation for microdontic teeth #12 and canine substitution #23. The periodontist and restorative dentist were also involved in planning and completing the treatment.

Prior to orthodontic treatment, an objective examination revealed adequate dentoalveolar development and dental arch growth in the patient. The cleft site, which had undergone previous surgical closure, appeared well healed with minimal scarring and no evidence of oronasal fistula. The right side of the cleft allowed the teeth to erupt properly, but the lateral incisor (#12) displayed microdontia, and the prolonged retention of the deciduous tooth (#53) delayed the eruption of the canine (#13). These findings indicated the presence of sufficient alveolar bone to support the eruption and movement of these teeth.

During the orthodontic treatment, the patient underwent several procedures to correct dental alignment, including levelling and unravelling. Initially, a fixed appliance pre-adjusted slot 0.022" Roth was bonded to the teeth, and a 0.022" Roth buccal tube slot was placed on teeth #16, #26, #36, and #46. The size of the archwire was gradually increased in both the upper and lower arches, starting with Niti 0.012", followed by 0.014", 0.016", and finally 0.018". Owing to its palatoversion and the coverage of its labial surface by tooth #11, the bracket for tooth #12 was not bonded at the beginning of the treatment. Once all teeth had been aligned, except for #12, an open coil spring was utilised to create space for teeth #12 and #13 on 0.016 × 0.022 stainless steel wire. In the lower jaw, the excess space was closed by distalising the teeth individually with ligatures in the posterior region, and a power chain was applied from the lower first molars to the second premolars. Furthermore, when there was enough space for teeth #12 and #13, tooth #12 was distalised to the ideal position. On the left side, the teeth were mesialised one by one to close the space due to the agenesis of tooth #22 using intra-arch elastic. Dental distalisation was continued in the lower jaw.

After the tooth #12 alignment was achieved, a bracket was bonded to tooth #13 to achieve the optimal curve. This procedure used a double-wire method, a combination of a primary wire of 0.016 × 0.022" stainless steel and an additional Niti 0.012". Ligatures are applied to the upper right posterior and anterior teeth to secure their positions and prevent any movement towards the space prepared for tooth #13. At the same time, mesialisation of the left maxillary teeth was carried out individually to close the gap caused by the agenesis of tooth #22 using intra-arch elastics. The anterior segment of the lower jaw was retracted. The final step was finishing and settling the occlusion using a 0.017 × 0.025" stainless steel archwire.

Radiographic evaluation using an orthopantomogram (Fig. 5a) revealed the presence of a permanent canine (#13) tooth bud in a vertical position, with plans for its traction. After 17 months of orthodontic treatment, the results were satisfactory, with tooth #13 successfully erupting into the ideal dental arch position. Secondary alveolar bone grafting in the cleft area was not performed because, based on the OPG examination after 17 months of treatment, radiopaque areas were observed around the apical region of tooth #13 (Fig. 5b), indicating adequate alveolar bone support surrounding its root.

After orthodontic treatment, the fixed appliances were debonded. Furthermore, the patient was referred to a conservative specialist dentist for veneer restoration, microdontia #22, and canine substitution tooth #23. A Hawley retainer was used for the maxillary and mandibular arches to maintain the teeth's position and for arch stabilisation.

Soft tissue analysis indicated that the upper and lower lip positions were more forward by the end of treatment (Table 1, Fig. 6). An improved profile and a good overjet, overbite, and interincisal relation were established. The upper and lower incisors were aligned in an optimal relationship (Table 1, Figs. 1c, 1d, 2c, 3c, 3d, 4b, 5b, 6).

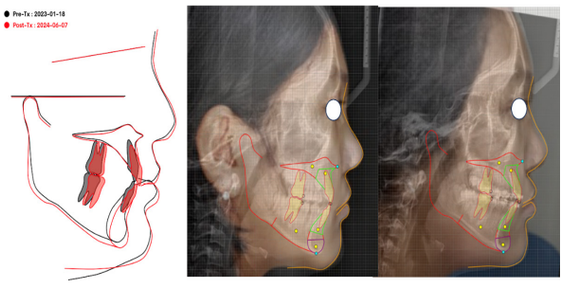

Fig. 6 Superimposition of lateral cephalometric.

DISCUSSION

The main objective of the CLP treatment is to anatomically close the defects to enhance feeding and speech development while avoiding any significant obstacle to maxillary growth or an increase in the prevalence of fistulas (Shkoukani *et al.*, 2013). The patient received her primary labioplasty when she was four months old, and then she underwent a palatoplasty procedure at the age of 17 months. This process aligns with the established guidelines, which recommend performing lip surgery on newborns between three and six months of age and palate surgery between 12 and 18 months. These procedures should ideally be completed before the child reaches 18 months, as this marks a critical stage in speech development (Rohrich *et al.*, 2000).

Orthodontic care for patients with CLP necessitates a collaborative multidisciplinary approach. The primary objective of the treatment was to achieve a more functional and aesthetically pleasing facial profile and dentition to enhance the patient's quality of life (Akbulut, 2020; Parsaei *et al.*, 2020; Kaiser *et al.*, 2024). Several stages in this treatment must be carried out. Patients with a CLP are more likely to have gingivitis, calculus, and dental caries than non-cleft patients. Risk factors include those related to the cleft condition or certain complications from subsequent surgical interventions. Malformations of the maxillary bone or teeth, poor dietary practices, and improper tooth brushing techniques in CLP patients can increase the susceptibility to periodontitis and dental caries (Wu *et al.*, 2023). Mouth preparation was performed on this patient before starting the orthodontic treatment. The initial steps involved scaling to enhance the health of the periodontal tissues, along with the extraction of residual roots from deciduous teeth that could potentially lead to infections in the oral cavity. The patient's motivation and cooperation are essential in attaining the treatment goals, and the parents must sign an informed consent acknowledging receipt of good information and agreeing to the course of treatment.

A comprehensive treatment plan that included straightwire orthodontic treatment and conservative dental strategies was suggested to attain normal function, proper occlusion, and a harmonious profile. Following the patient's panoramic radiographic examination for orthodontic treatment, a cleft was observed in the alveolar ridge between tooth #12 and persistent tooth #53 (Fig. 5a). After the extraction of tooth #53, a passive open coil spring was used to keep the space until tooth #13 successfully erupts. The bracket position at tooth #13 was adapted to move tooth #13 carefully into the dental arch, avoiding moving the root into the cleft area. Although the patient has no prior history of bone grafting in the cleft area, by the end of the treatment, the cleft appeared favourable, with tooth #13 successfully aligned within the dental arch (Fig. 5b).

The orthodontic requirements of patients presenting with UCLP/BCLP may differ based on the severity of the existing

dentofacial deformities. In this case, the patient experienced microdontia #12, agenesis #22, anterior crossbite #11 to #42, and #13 to #43, as well as several malpositioned teeth. Common dental anomalies in patients with an orofacial cleft include tooth shape differences like microdontia and hypoplastic enamel, agenesis, impacted or ectopic teeth, and transpositions. A deficiency in the vascular supply and mesenchymal support near the cleft site frequently leads to the congenital absence of maxillary lateral incisors. Patients with UCLP also exhibit a constricted maxillary arch and anterior crossbite, which may occur with or without a posterior crossbite on the cleft-affected side (Akbulut, 2020; Parsaei *et al.*, 2020; Paradowska-Stolarz *et al.*, 2022).

Orthodontic treatment aims to correct the malrelationship and malposition of individual teeth and to align the dental arch to achieve good occlusion. The outcomes of orthodontic intervention in this patient's case are satisfactory and the results were achieved within a comparatively brief period (17 months). The patient's age was favourable for the success of orthodontic treatment. Adolescence is identified as a critical period for orthodontic treatment. The best period is when young children enter the pubertal growth spurt, and the body grows rapidly until the growth and development are completed. This age range allows for correcting malocclusions as the jaw and teeth are still developing, making it easier to achieve desired outcomes (Alhasyimi & Syahfik, 2023; Zhou *et al.*, 2024). In the final phase of treatment, to obtain optimal treatment results, patients are referred to a conservative dentistry specialist to carry out dental veneer restorations of teeth #12 and #23 to achieve a more esthetic result.

After orthodontic treatment and conservation are completed, retainers are required to maintain stability. The selection of retainers may differ based on the specific case (Kim *et al.*, 2021). Especially the degree of scarring of the palate should be considered. If there is heavy scarring with clear scar bands along the alveolar process and across the palate, it will be difficult to maintain the stability of the transverse dimension (Kuijpers-Jagtman & Kuijpers, 2023). A recently published, evidence-based clinical practice guideline regarding CLP advocates retaining the upper front teeth with a fixed retainer bonded to all anterior teeth. Furthermore, it recommends the utilisation of a removable orthodontic retainer, such as a Hawley retainer, to uphold the maxillary transverse dimensions. Such a retainer ought to be worn nightly lifelong (Mink van der Molen *et al.*, 2021).

The limitation in this case is trying to achieve an optimal occlusion with Canine Class I on the right side due to the difference in the number of teeth in the upper and lower jaws. Nonetheless, in terms of aesthetics and functionality, this is the optimal outcome.

CONCLUSION

The orthodontic management of UCLP associated with dental agenesis and microdontia has presented considerable challenges. Implementing a multidisciplinary approach has resulted in substantial enhancements to facial and dental aesthetics and functional capabilities. The patient had assessed the results as satisfactory.

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