Multidisciplinary treatment from infancy to adolescence of a patient with a unilateral cleft lip and palate: a 16-year follow-up case report

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Cleft lip and palate (CLP) are conditions that require long-term management and review from infancy to adolescence. Surgical procedures become easier when nasoalveolar moulding (NAM) measures are applied during the neonatal period. Orthodontic treatment provided for these patients during the progressive dentition developmental periods can achieve positive aesthetic and functional results. In patients affected by a CLP, the lateral incisor in the cleft region is usually missing and multidisciplinary prosthodontic rehabilitation of the edentulous space is often required. In the present case report, positive results of NAM, coupled with orthodontic and prosthodontic treatment are presented for a CLP patient who began management during the neonatal period. In addition, a minimally invasive aesthetic restoration is presented as a solution for the prosthodontic rehabilitation of a missing lateral incisor.

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Introduction

As the most common craniofacial anomaly, a patient born with a cleft lip and/or palate (CL/P), presents a complex disorder. CL/P can be "syndromic," occurring as, or a part of a syndrome, as well as occurring in isolation or "non-syndromic."^{1,2} Although its aetiology is often not fully known, CL/P is a congenital deformity associated with hereditary and environmental factors seen in approximately one in 1000 live births.^{3,4} It is characterised by ethnic and geographical differences which often negatively impact on an individual's psychological state by causing an aesthetically challenging appearance. Furthermore, the functions of speaking and mastication are affected, which causes additional problems related to the integrity of the patient's stomatognathic system. Therefore, CL/P management requires a multidisciplinary approach to meet the significant needs of the patient.^{4,5} The multidisciplinary approach usually involves a long process starting from infancy and extending into adulthood. 4,6

Although deformities involving CL/P occur at different levels and severities, infant orthopaedic treatment, usually applied soon after birth, has been advocated since the 1950s.^{7,8} Using nasoalveolar moulding (NAM) during early infancy, orthodontists bring the cleft lip, alveolar and palatal segments closer together, to facilitate the surgeon's ability to close the primary lip defect as well as improve feeding of the cleft child.⁹ Surgical treatment for cleft patients covers a period between the first 6 months and 2 years after birth but is usually completed around 18 months, at a time when speech development typically begins.

If orthodontic treatment is indicated for non-cleft patients with missing teeth, spaces are preferably closed. The advantages include a lower cost, fewer procedures, enhanced aesthetics, and simpler life-long dental care for the patient.¹⁰ However, in patients with an alveolar cleft, orthodontic space closure is not possible due to a lack of bone in the area of the agenesis.^{11,12} Therefore, prosthodontic rehabilitation of missing teeth is recommended as part of the multidisciplinary approach.¹³ Traditionally, anterior fixed restorations are used in CL/P patients;^{5,14–16} however, implant-supported prosthodontic applications can provide a minimally invasive treatment alternative.¹⁷ Bone grafting and additional surgical procedures are required for implant placement in CL/P patients^{12,18} but, if restoration is attempted at too early an age, aesthetic and functional complications related to growth and development, arise.¹⁹

To evaluate the appropriateness of a multidisciplinary treatment program, the following clinical case describes a patient from infancy into adolescence (1 week to 16 years old) who presented with a left unilateral complete cleft lip and palate (UCLP). The report indicates that aesthetically acceptable results can be obtained in affected patients by comprehensive treatment performed at the appropriate time using current minimal prosthetic methods.

for presurgical nasoalveolar moulding. The patient's parents requested treatment due to the poor aesthetics caused by the CLP and the nasal regurgitation of food. On presentation, there was a 6–7-mm-wide gap between the cleft alveolar segments. The morphology of the nose and base on the cleft side was distorted due to flattening of the nasal wing (Figure 1A).

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Two treatment options were presented to the patient/ parents following the initial clinical examination. The first involved closure of the cleft by a direct surgical procedure but this method did not permit the shaping of the nasal cartilage and the level of tension in the tissues during gingivoperioplasty produces aesthetically inadequate results. A nasoalveolar moulding technique, allows the shaping of the nasal cartilage, reduces tension by approximating the lip components, provides better bone formation with a reduced alveolar cleft dimension, and reduces the need for a secondary bone graft. This was presented as a second treatment option and accepted by the patient's parents.9,20-22 The nasoalveolar moulding was performed following the method suggested by Grayson.^{9,20} In addition to pretreatment dental casts and photographic records, a further maxillary impression was taken of the patient using C-type silicone impression material (Zetaplus C-Silicone Impression Material, Zhermack, Badia Polesine, Italy). An emergency medical team supported the

Case report

A 1-week-old female patient, born with a complete left side UCLP (Figure 1A) was referred to the



Figure 1. (A) When the patient applied to the clinic in the neonatal period. (B) Appearance of the appliance used for NAM before surgery. (C) Final state before lip surgery.

patient in case a problem was encountered during the impression procedure. The NAM appliance was made using acrylic resin (Orthocryl, Dentaurum, Ispringen, Germany) (Figure 1B) and the material thickness was set at 2–3 mm for structural integrity.²³ The appliance was worn for 3 months, and weekly reviews were conducted.²⁴ During the review visits, adhesive lip strips were applied to approximate the cleft segments. Roughening of the acrylic and the addition of an inner soft lining material helped cushion and guide the cleft alveolar components. At the end of the moulding process, the distance between the alveolar cleft was considerably reduced (Figure 1C). Subsequently, the patient's lip was surgically closed and repaired. Upon re-examination when the patient was 2 years old, the cleft palate was surgically treated (Figure 2).

The patient was further re-examined during the mixed dentition period (8 years old; Figure 3) at a time when the patient complained of missing teeth in the anterior region, caries, and an inability to pronounce words correctly. When the patient reached the age of 12, an assessment for fixed orthodontic treatment

was conducted (Figure 4). Clinical examination findings were a maxillary narrowness (Figure 4), an asymmetric smile, moderate crowding in both arches, a lower midline deviation to the left, an upper midline deviation to the right, the presence of the mandibular right and left primary second molars, agenesis of the maxillary left lateral incisor in association with the cleft defect (Figure 4). The radiographic findings confirmed the absence of the upper permanent left lateral incisor, and the upper permanent left canine was attempting to erupt (Figure 4). A cephalometric analysis revealed a skeletal class I malocclusion (ANB: 1.6°), A-Nperp (-2.5 mm) and Pg-Nperp (-6.8 mm) measurements showed the lower and upper skeletal bases were retrognathic, but with an acceptable vertical relationship (SN/GoGn: 36.0). The upper incisors were retroclined (U1/SN: 97.1°; U1/PP: 106.5°; U1/ NA: 21.2°) and retruded (U1-NA: 2.7 mm). The lower incisors were in a normal position (L1-APog: 2.0 mm; L1-NB: 3.1 mm) and inclination (IMPA: 90.1°; L1/NB: 23.0°), but with a -1.6 mm overjet and 1.4 mm overbite (Table I). The patient was presented



Figure 2. Follow-up records at age 2.

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Figure 3. Front view of lips, nose, and mouth at 8 years old.



Figure 4. Photographs of the patient before and during orthodontic treatment in the mixed dentition period. Pre-treatment intraoral and extraoral profile and frontal photographs and panoramic and cephalometric radiographs.

Measurements	Before treatment	End of treatment	6 months follow-up	2 years follow-up
SNA°	75.8	76.2	76.3	76.3
SNB°	74.3	74.8	74.1	74.5
ANB°	1.6	2.4	2.2	1.8
A-Na Perp (mm)	-2.5	-2.2	-1.7	-1.1
Pg-Na Perp (mm)	-6.8	-5.6	-4.6	-2.4
ANS-Me (mm)	55.6	59.4	59.5	59.6
SN/GoGn°	36.0	37.9	36.3	35.7
NaMe (AFH) (mm)	103.1	109.8	110.5	111.4
SGo (PFH) (mm)	62.8	67.7	69.1	70.0
N-ANS (UFH) (mm)	48.3	51.9	51.5	52.3
ANS-Gn (LFH) (mm)	54.0	59.2	58.5	58.6
N-Gn (TFH) (mm)	102.3	111.1	110.0	110.9
CoA (mm)	75.3	79.1	80.1	80.6
CoGn (mm)	101.0	108.1	108.9	109.2
U1/SN°	97.1	104.3	106.6	106.8
U1/PP°	106.5	115.6	119.2	122.8
U1-NA (mm)	2.7	4.8	4.9	6.4
U1/NA°	21.2	28.1	29.4	30.9
L1-APog (mm)	2.0	3.4	4.0	4.8
IMPA°	90.6	97.8	99.7	100.3
L1-NB (mm)	3.1	6.0	6.7	6.9
L1/NB°	23.0	32.0	33.0	33.1
/ °	134.3	116.5	113.9	114.5
Overbite (mm)	-1.6	1.5	0.9	0.8
Overjet (mm)	1.4	3.3	2.5	2.1
NLA°	91.4	84.1	83.9	80.4
LL-E (mm)	-0.3	-0.1	0.7	2.3
UL-E (mm)	-0.7	-1.0	-0.7	-2.2
Soft Tissue Convexity°	133.7	136.5	135.6	137.1

Table I. Cephalometric measurements of the patient who was treated and followed up

^oDegree; (mm): millimeter; SNA: Sella-Nasion-A point; SNB: Sella-Nasion-B point; ANB: A point-Nasion-B point; A-Na Perp: Na Perpendicular to A point; B-Na Perp: Na Perpendicular to B point; ANS-Me: Anterior Nasal Spine-Menton; SN/GoGn: Sella-Nasion/ Gonion-Gnathion; NaMe (AFH): Nasion-Menton (Anterior Facial Height; SGo (PFH): Sella-Gonion (Posterior Facial Height); N-ANS (UFH): Nasion-Anterior Nasal Spine (Upper Facial Height); ANS-Gn (LFH): Anterior Nasal Spine-Gnathion (Lower Facial Height); N-ANS (UFH): Nasion-Gnathion (Total Facial Height); CoA: Condylion-A point; CoGn: Condylion-Gnathion; U1/SN: Long Axis of Upper Central Incisor/ Sella-Nasion; U1/PP: Long Axis of Central Upper Incisor/Palatale Plane; U1-NA: Perpendicular distance measured from the tip of the upper central incisor to the Nasion-A line; U1/NA: Long Axis of Central Upper Incisor/Nasion-A point; L1-APog: Perpendicular distance measured from the tip of the lower central incisor to the Pogonion-A point; IMPA: Long axis of lower central incisor/Mandibular Plane; L1-NB: Perpendicular distance measured from the tip of the lower central incisor to the Nasion-A line; L1/NB: Long Axis of Lower Central Incisor/ Nasion-B point; I/I: Interincisal Angle; NLA: Nasolabial Angle; LLE: Lower lip-Ricketts E Line; UL-E: Upper lip-Ricketts E Line.

with three treatment alternatives: (1) treatment that would begin immediately, and involve the application of fixed orthodontic appliances involving expansion of the narrow maxilla, followed by the application of a retention regime that would incorporate the missing lateral incisor; (2) commencement of the patient's orthodontic treatment later in adulthood, followed by a fixed prosthetic restoration of the lateral incisor; (3) no treatment if neither of the options were preferred.

The parents consented to option 1 and planned treatment included rapid maxillary expansion and the use of a lingual arch until the exfoliation of the mandibular primary second molars, before the placement of fixed appliances. Prior to the expansion procedure, bone grafting was recommended to the family, but it was rejected due to the perceived negative psychological effects of the surgery. The expansion appliance was designed using a single 10 mm screw (Dentaurum, Ispringen, Germany), in which all teeth and palatal tissues in the posterior region were covered with acrylic resin (Imicryl, Konya, Turkey), but leaving the upper permanent canines free in the anterior region. Activation of the cemented appliance was performed twice a day (0.2 mm per activation), until the palatal cusps of the upper first permanent molars were aligned with the buccal cusps of the lower first permanent molars.²⁵ After the achieved expansion, bands were cemented to the maxillary permanent first molars, and brackets (Mini Master; American Orthodontics, Sheboygan, WI, USA) and arch wires were applied between a custom-made transpalatal arch and the maxillary canines for anchorage (Figure 5). In co-ordination with a prosthodontist (FY), future prosthetic rehabilitation was planned to meet aesthetic and functional requirements by replacing the agenic lateral incisor.

The orthodontic treatment extended over 2 years and 3 months at which time a dental class I molar and canine relationship was achieved along with an appropriate overjet and overbite (Figure 6). According to the cephalometric comparison at the end of treatment, the ANB angle increased as predicted (2.4°), the A-Nperp (-2.2 mm) and Pg-Nperp (-5.6 mm) values decreased, the vertical dimensions of the face increased (Table I), the maxillary and mandibular teeth improved to protruded and proclined positions, the interincisal angle decreased, the overjet and overbite values increased, the nasolabial angle decreased, and the soft tissue convexity increased (Table I). As a result of the well-established occlusion and an acceptable dentofacial profile, the fixed orthodontic treatment avoided the need for orthognathic surgery required by many CLP patients. All cephalometric tracings and analyses were performed using Dolphin



Figure 5. Appearance of anterior brackets and custom-made transpalatal arch applied after maxillary expansion procedure in the maxilla and lingual arch in the mandible.

MULTIDISCIPLINARY CLEFT LIP AND PALATE TREATMENT

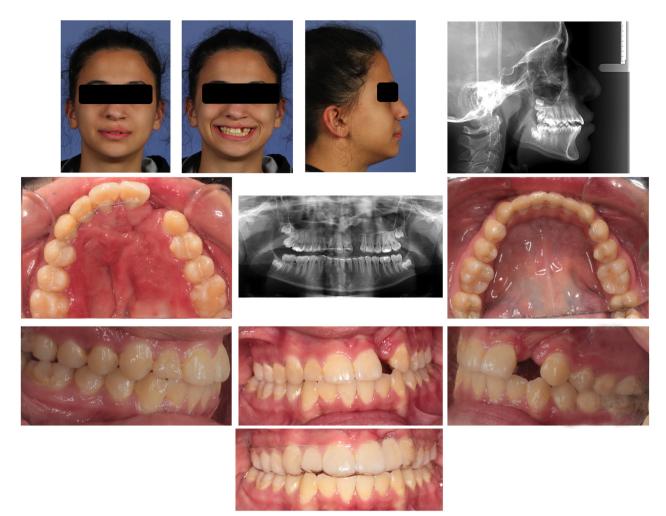


Figure 6. Post-treatment intraoral and extraoral profile and frontal photographs and panoramic and cephalometric radiographs. Modified essix retainer with acrylic artificial teeth applied for retention after orthodontic treatment.

Imaging software (Version 11.0; Dolphin Imaging and Management Solutions, Chatsworth, CA, USA). Monthly oral hygiene motivation was provided to the patient to manage white spot lesions which formed during treatment. Toothpaste and mouthwashes containing fluoride were suggested, and a low-cariogenic diet was recommended to prevent further lesions. However, the patient's motivation and co-operation fluctuated during active treatment and oral hygiene adherence changed from good to moderately poor. At the end of treatment, white spot lesions were noted, and the patient was provided with continuing oral hygiene support (Figure 6).

The crowding in the lower and upper dental arches was resolved, and the dental midlines were relatively corrected. Finally, following a prosthodontic consultation, space was opened for the missing maxillary lateral incisor to enable future multidisciplinary treatment. Orthodontic retention was provided by a fixed lingual retainer (Reliance, Itasca, IL, USA) attached using flowable light-cure composite resin (Filtek Supreme XTE, 3M, St. Paul, MN, USA) between the first premolars in the lower arch (Figure 3B, D) and the right canine and left central incisor in the upper arch (Figure 6).

When growth and development were completed, the patient was presented with either a dental implant or a fixed prosthetic restoration as two options that would be aesthetically acceptable. However, until an appropriate age was reached, the replacement alternatives were the use of a removable appliance incorporating an acrylic tooth or the application of a fixed modified resin-bonded bridge restoration. The patient preferred the use of a removable appliance (Figure 6) and was reviewed 6 months after appliance insertion. At this time the patient stated that the appliance had not been worn and requested a fixed solution. The patient accepted a fixed retention alternative in the form of a bonded artificial tooth. For financial considerations at the age of 16 years, the patient was provided with a modified Maryland prosthetic bridge (Figure 7) in which an acrylic tooth was incorporated into a multi-stranded lingual retainer²⁴ (Reliance, Itasca, IL, USA). In a further review conducted 2 years and 6 months later, it was noted that occlusal stability had been maintained, while the patient's growth continued (Figure 8). After an additional 6 months, it was determined that the prosthetic restoration was functional and stable.

Discussion

The shape of the face in the early stages of embryonic development depends on the relationship between the genes involved in organogenesis and the tissues that make up the face. A disruption of the relationship leads to incomplete fusion and the development of a cleft of the face, lips and/or palate.^{26,27} A cleft lip and/or palate (CL/P) is the most common example of a facial cleft and a leading congenital craniofacial deformity.²⁷ The parents of infants with a CL/P are usually made aware of the condition before birth, through routine ultrasound checks during pregnancy.^{27,28}

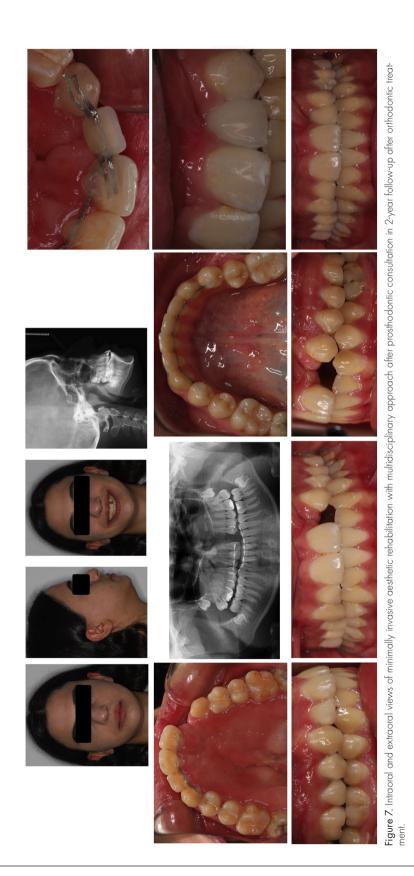
The present case report demonstrates that presurgical maxillary orthopaedic treatment performed in accordance with NAM procedures, together with a long-term review, produces positive treatment results.^{29,30} Although the encouraging effects of NAM in patients affected by a unilateral cleft lip and palate (UCLP) have been described by Padovano et al.,³⁰ the long-term effects of the procedure are unclear. The additional care burden and frequent appointments create challenges, and alternative procedures such as DynaCleft (Southmedic, Ontario, Canada) have been established when traditional presurgical orthopaedic treatment is inappropriate.³¹

The present case report demonstrates that presurgical maxillary orthopaedic treatment performed in accordance with the NAM protocol, together with long-term review, produces positive results. As noted by Matsuo et al.,³² the first 6 weeks after birth provide a window of opportunity to facilitate CLP surgery by taking advantage of the extreme plasticity of the neonate tissues. Matsuo et al.³² shaped the alar cartilage and brought the cleft lip and alveolar segments closer together, as recommended by Grayson et al.^{9,20} A 3-month follow-up was performed on the newborn, at which time, lip surgery has been recommended.^{20,33}

The current patient, who was reviewed for an extended time, was assessed for orthodontic treatment in the late mixed dentition period when the use of fixed appliance mechanics was accepted. As previously identified by Perillo et al.,³⁴ the presented CLP patient had a complex list of problems related to significant maxillary narrowness, a missing maxillary left lateral incisor, crowding in both arches, irregularity of the dental midlines, and a decreased overbite-overjet relationship. Additionally, there was no space for the maxillary right canine, and the left canine was erupting ectopically. At the outset, a rapid maxillary expansion appliance was applied for maxillary transverse correction.^{25,34-36} However, Ramstad and Jendal³⁷ found a significant decrease in maxillary transverse dimension after the removal of the appliance in UCLP patients. Therefore to preserve stability, fixed and removable retainers in combination were inserted after treatment. No relapse was subsequently observed during the followup periods of 6 months and 2 years, respectively. Fixed orthodontic treatment procedures were later provided and appropriate dental and skeletal treatment was performed.34 As a result of the patient's continued growth and development and the effects of orthodontic treatment, an acceptable correction was achieved in lower and upper incisor positions to suggest an improvement in the maxillomandibular relationship. It was expected that a significant ANB change would be caused by a change in the SNA angle and, as a result of this change, it was considered that the patient did not need orthognathic surgery. In addition, the ANB change produced a clinically acceptable profile.³⁸

Several previous studies have applied multidisciplinary treatment procedures and careful consideration^{5,11,14} to manage CLP patients.^{4–6,13,34–36} Because of a lack of appropriate options to manage the anterior spaces found in CLP individuals,^{11,39} the prosthodontic rehabilitation of affected patients presents significant challenges. In cases involving missing lateral incisors, it is unwise to move the permanent canine to an adjacent position relative to the central incisor due to the lack of alveolar bone.¹² Furthermore, because of the aesthetic morphologic and color limitations of the canine, placing those teeth adjacent to the central incisor is not preferred.^{11,40,41}

Minimally invasive aesthetic procedures to manage the absence of the lateral incisors have been reported



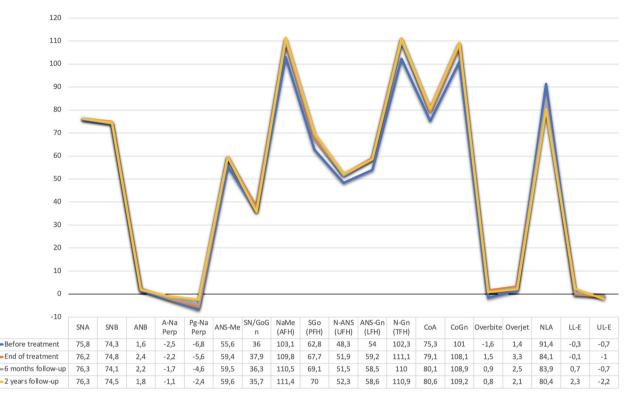


Figure 8. Graphical representation of cephalometric changes that occurred because of orthodontic treatment and subsequent growth and development.

in recent studies.^{11,42} In the treatment of the present case, the missing lateral incisor was to be managed by a procedure promoted by Kravitz.42 However, a simple and economical treatment alternative using fewer materials was offered, which is especially useful in countries of low socioeconomic status. In addition, unlike the minimally invasive aesthetic procedure recommended by Malgaj et al.,11 the management protocol included a more restorative approach on healthy teeth adjacent to the missing tooth and can be clinically applied without the need for a rubber-dam. Compared to a case reported by Szmidt et al.,¹⁴ the applied modified Maryland bridge procedure incorporated fewer restorative demands on teeth adjacent to the cleft, although both options allowed treatment of the patient in a single appointment.

At the end of treatment, the initial goals were achieved with adequate occlusal, functional and aesthetic results despite the lack of grafting in the maxillary left lateral incisor region and a lack of co-operation. Although the use of ceramic adhesive bridges, which was not preferred by the patient, could have produced a superior aesthetic outcome, the result was still clinically satisfactory particularly for patients and their parents who demand a stable result. Multidisciplinary (orthodontics and prosthodontics) co-operation and the patient's motivational efforts were effective in ensuring a successful overall treatment outcome. By this application, the cleft deformity, and its greater psychological effects in adolescence compared to pre-adolescence,⁴³ was effectively treated.

Conclusion

The treatment of CLP, as a common dentofacial anomaly characterised by maxillary narrowness, missing teeth and impaired aesthetics, requires complicated and extensive treatment.

As the present case report demonstrates, positive functional and aesthetic results can be obtained by monitoring a UCLP patient from infancy by using appropriate and accepted multidisciplinary techniques. The minimally invasive procedures and aesthetic prosthodontic restoration applied in this case provided adequate aesthetic results in a patient who refused a bone graft. In a patient who is not suitable for an implant, it is suggested that the current protocol can be considered for the prosthodontic rehabilitation of a CLP patient.

Conflict of Interest

The authors declare that there is no conflict of interest.

Informed consent

A written informed consent form was obtained from the patient's parents before all treatment procedures.

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