Interdisciplinary treatment of a patient with bilateral cleft lip and palate and congenitally missing and transposed teeth

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The comprehensive treatment of a patient with cleft lip and palate requires an interdisciplinary approach for functional and esthetic outcomes. A 20-year-old woman with bilateral cleft lip and palate had a chief complaint of unesthetic appearance of her teeth and the presence of oronasal fistulae. Her clinical and radiographic evaluation showed a dolichofacial growth pattern, a Class II skeletal relationship with retroclined maxillary central incisors, 5 mm of negative overjet, maxillary constriction, maxillary and mandibular crowding, congenitally missing maxillary right incisors and left lateral incisor, and a transposed maxillary left canine. Her treatment plan included the extraction of 3 premolars, maxillary expansion, segmental maxillary osteotomy, repair of the oronasal fistulae, rhinoplasty, periodontal surgery, and prosthodontic rehabilitation. To obtain a better occlusion and reduce the dimensions of the fistulae, orthognathic surgery comprising linear and rotational movements of the maxillary segments (premaxilla, right and left maxillary alveolar segments) in all 3 axes was planned by performing 3-dimensional virtual surgery on 3-dimensional computerized tomography. At the end of the interdisciplinary treatment, a functional occlusion, a harmonious profile, and patient satisfaction were achieved. Posttreatment records after 1 year showed stable results. (Am J Orthod Dentofacial Orthop 2014;145:381-92)

Cleft lip and palate (CLP) is a congenital deformity that is associated with maxillary sagittal, transversal, and vertical discrepancies.[1,2] In addition to skeletal discrepancies, this deformity is often accompanied by dental abnormalities, such as hypodontia, hyperdontia, and transpositions. Hypodontia, especially the absence of the maxillary lateral incisors, is the most prevalent.[3,4] The combination of the skeletal and dental malocclusion with the soft-tissue and hard-tissue deformities or deficiencies complicates the treatment of CLP and requires interdisciplinary approaches to obtain functional and esthetic outcomes.

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In recent years, 3-dimensional (3D) virtual planning of orthognathic surgery has begun to be used in clinical practice.[5-9] Computer-aided surgical simulation enables doctors to perform osteotomies, reposition the osteotomized bony structures, control intercuspation, control interferences between osteotomized bony structures, evaluate difficulties before surgery, and simulate the postoperative results on the hard and soft tissues in 3 dimensions. The challenging treatment of CLP can benefit from 3D surgical planning because of the complex characteristics of the malocclusion and the patient's unique and individual requirements. In patients with alveolar clefts in whom integrity of the alveolar segments was not achieved, treatment becomes multipiece maxillary segmentation after LeFort I osteotomy. When multipiece maxillary segmentation is used, it is not possible to describe the intraoperative movements of each maxillary segment with conventional surgical planning. Computer-aided surgical simulations have advantages, especially for these patients.

The aim of this case report was to present the interdisciplinary treatment of a 20-year-old woman with bilateral CLP and congenitally missing and transposed teeth.

DIAGNOSIS AND ETIOLOGY

A 20-year-old woman with operated nonsyndromic bilateral CLP was referred to the orthodontic clinic of

Fig 1. Pretreatment facial and intraoral photographs.

Yeditepe University in Istanbul, Turkey. Her chief complaints were the unesthetic appearance of her teeth and the presence of oronasal fistulae. She had received primary lip repair and palatoplasty in the first year of life and did not undergo bone grafting. Her extraoral examination showed a slight asymmetry at the eye level but no apparent mandibular asymmetry. She had nasal deviation, widening of alar bases, and loss of columellar projection. Her retruded upper lip was improperly repaired and scarred. Her lower lip was protruded. Her V-shaped maxillary arch was severely constricted. The premaxillary segment was mobile. Her intraoral photographs showed anterior and posterior bilateral crossbites, severe maxillary and mandibular crowding, a deep curve of Spee, Angle Class II molar relationships, a palatally displaced and rotated left second premolar, a transposed maxillary left canine, and a hypomineralized maxillary central incisor. The maxillary left deciduous canine had extensive caries. Bilateral alveolar fistulae were present (Figs 1 and 2).

The panoramic radiologic evaluation showed that the patient had congenitally missing maxillary right incisors and the left lateral incisor, and residual maxillary left deciduous lateral incisor and canine. Osseous defects were apparent bilaterally, and there was a small amount of osseous support of the central incisor in the premaxillary segment (Fig 3, *A*). The cephalometric analysis showed a dolichofacial growth pattern, a Class II skeletal relationship, retroclined maxillary and mandibular central incisors, 5 mm of negative overjet, and a 1-mm overbite. The upper and lower lip to E-plane distances were −5.5 and 2.1 mm, respectively (Fig 3, *B*; Table).

TREATMENT OBJECTIVES AND ALTERNATIVES

The treatment objectives were to expand the maxillary arch; solve the maxillary and mandibular crowding; level the curve of Spee; obtain normal overjet, overbite, and incisor inclinations; correct the sagittal and vertical skeletal discrepancies by orthognathic surgery; close the

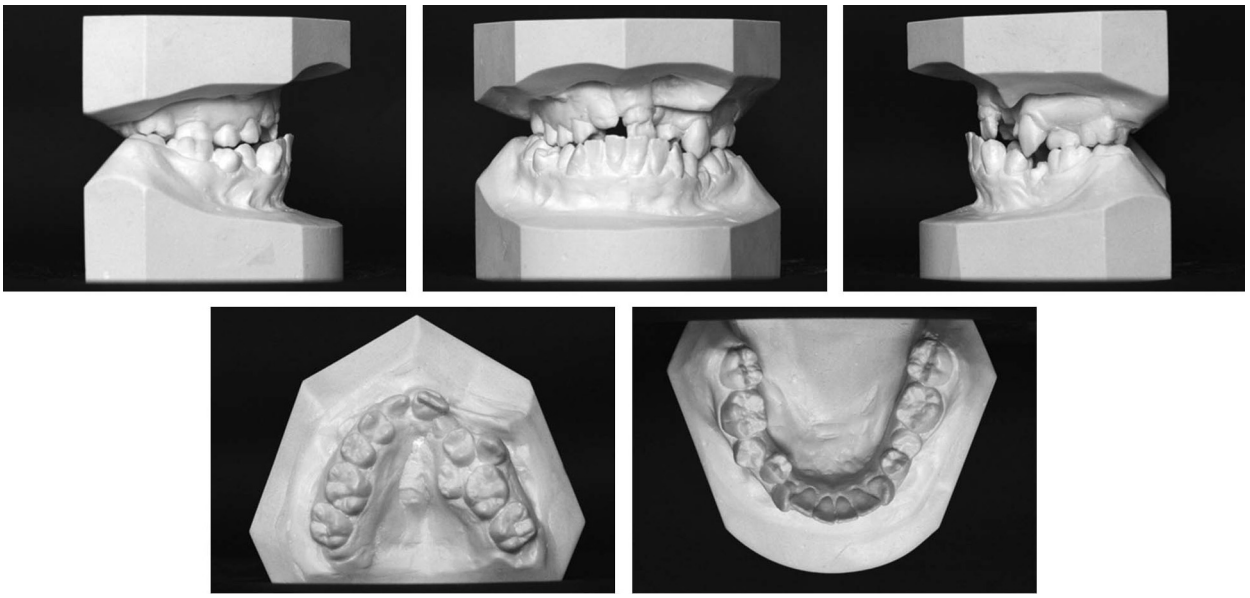

Fig 2. Pretreatment dental casts.

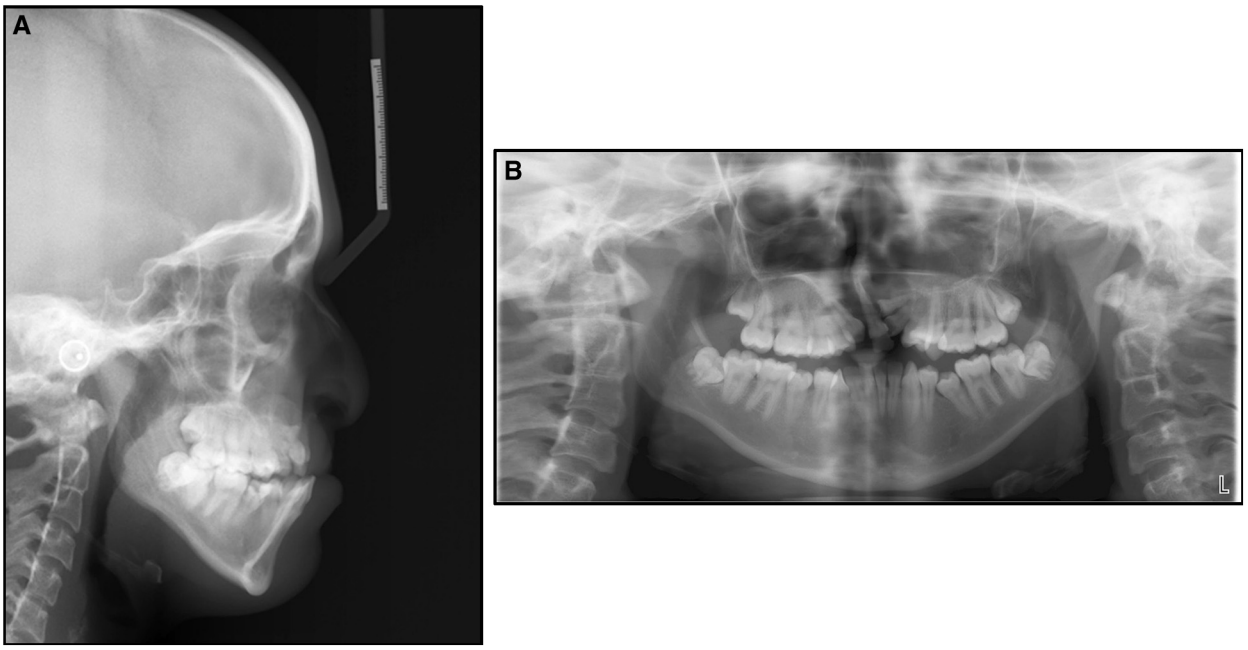

Fig 3. Pretreatment radiographs: **A,** lateral cephalometric radiograph; **B,** panoramic radiograph.

palatal osseous defects and oronasal fistulae; stabilize the maxillary arch using autogenous bone grafting; improve the facial profile; and rehabilitate the missing, transposed, and malformed maxillary teeth with an interdisciplinary approach by a plastic and reconstructive surgeon, an orthodontist, a prosthodontist, and a periodontist.

Several treatment alternatives were discussed by the team members and proposed to the patient regarding her main problems. For correction of the maxillary arch constriction, 2 options (expansion with an orthodontic appliance and expansion during multipiece maxillary surgery) were evaluated. Because of the palatal scar tissue, it was decided to avoid excessive transversal

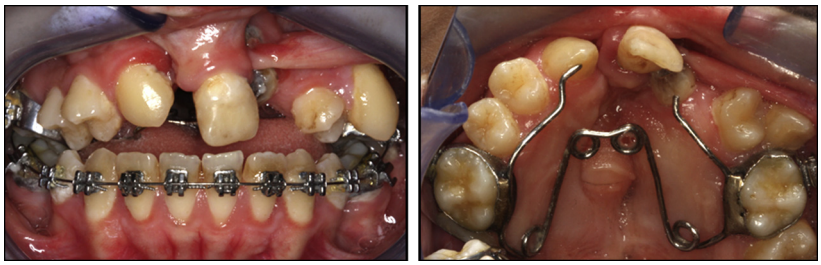

Fig 4. Intraoral photographs after expansion with the quad-helix.

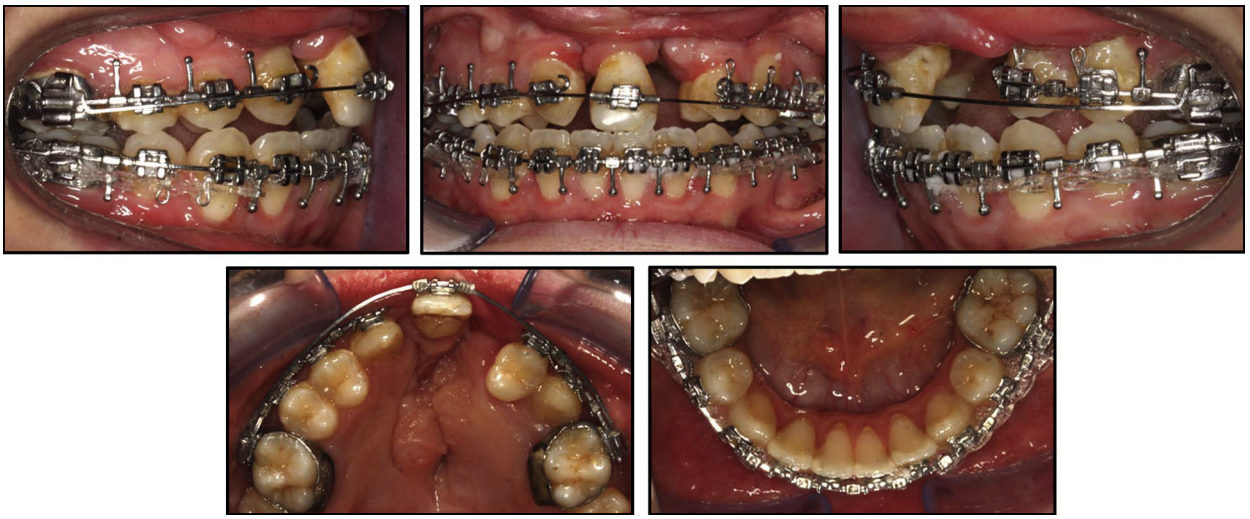

Fig 5. Preoperative intraoral photographs.

movements of the maxillary segments during orthognathic surgery. Therefore, orthodontic expansion was chosen. In case of insufficient expansion, surgical assistance would be required.

For the closure of the bilateral clefts and stabilization of the maxillary arch, autogenous bone grafting of the bilateral alveolar defects and repair of the oronasal fistulae with local flaps after expansion were considered. Alveolar distraction, especially to diminish the width of the clefts, was also proposed as an alternative.

Bimaxillary orthognathic surgery was considered first to correct the excessive lower facial height and the mandibular retrognathism, by impaction of the posterior maxillary segments and mandibular counterclockwise rotation. When this was discussed by the team, stability of the maxillary segments was thought to be better with maxillary surgery only, compared with double-jaw surgery. For leveling of the maxillary segments in all 3 axes, a LeFort I osteotomy with multiple bone segments was planned.

After the orthodontic-orthognathic treatment, prosthetic rehabilitation was planned. Various prosthodontic treatment options are available for patients with multiple missing teeth at the cleft area depending on the number and location of the missing teeth, the presence of sufficient alveolar bone and gingival tissues, and the stability of the maxillary segments, including dental implants and tooth-supported or implant-supported fixed or removable dentures.

For the improvement of her facial appearance, nose and lip revisions were also planned.

Our treatment objectives and alternatives were explained to the patient. She did not want to undergo mandibular surgery. She also declined the alveolar bone grafting because of morbidity of the donor site. Therefore, the final treatment plan comprised extraction of 3 second premolars, deciduous teeth, and third molars; maintenance of the positions of the transposed teeth; expansion; only maxillary surgery with multiple bone segments; repair of the oronasal fistulae; rhinoplasty; periodontal surgery; and prosthodontic rehabilitation.

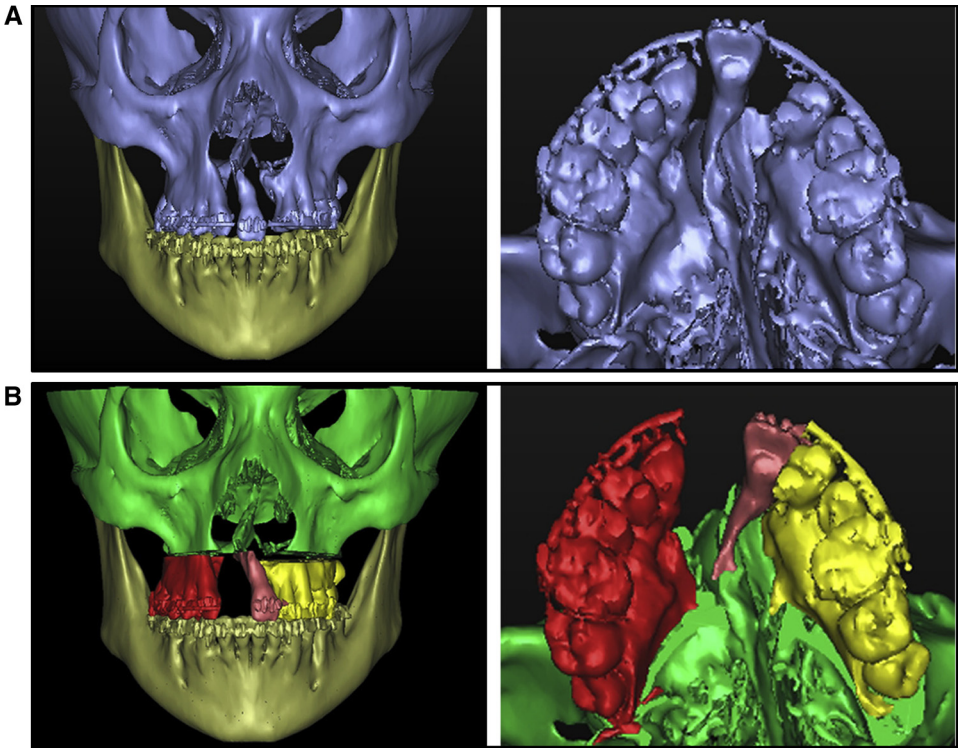

Fig 6. A, Preoperative 3D images; **B,** 3D surgical simulation and plan. Posterior movement of the right lateral segment, anterior movement of the left lateral segment, and distal repositioning of the premaxillary segment reduced the dimension of left cleft width.

TREATMENT PROGRESS

After extraction of the maxillary left second premolar and the mandibular second premolars, expansion of the maxillary dental arch was achieved with a quad-helix appliance. Expansion led to buccal tipping of the posterior segments and the posterior teeth, and opening of the bite (Fig 4). The maxillary posterior teeth and the mandibular dental arch were leveled and aligned using segmental and continuous mechanics, respectively, with 0.022-in fixed appliances (OmniArch; GAC International, Bohemia, NY). The mandibular extraction spaces were closed with sliding mechanics and minimum anchorage. The negative overjet and the inclination of the maxillary central incisor were corrected with proclination of the incisor. The position of the incisor was maintained with a 0.19 × 0.25-in stainless steel wire before surgery (Fig 5).

A computed tomography scan was obtained for orthognathic surgery planning. Before orthognathic surgery, the fistulae were further enlarged because of the lateral expansion and proclination of the premaxilla. All possible maxillary segment reposition scenarios were considered with virtual 3D orthognathic surgery software (Simplant OMS; Materialize Dental, Leuven, Belgium) to obtain a better occlusion and reduce the dimensions of the fistulae. The maxillary segments (premaxilla, right and left alveolar segments) were moved not only linearly but also rotationally in all 3 axes. The first surgical plan comprised achieving Class I molar relationships on both sides; this led to overexpansion of the clefts and the fistulae. In the second plan, the maxillary right segment was moved 1.8 mm posteriorly, 2.5 mm laterally, and 0.6 mm vertically, and it was rotated by −1.7°, −0.4°, and 7° on the x, y, and z axes, respectively, achieving a Class I molar relationship, whereas the left segment was moved 4 mm anteriorly, 2 mm laterally, and 2 mm vertically, and it was rotated by −6.8°, −2.1°, and 0.4° on the x, y, and z axes, respectively, to a Class II molar relationship. The maxillary left central incisor and the premaxillary segment were positioned 2 mm distally to correct the maxillary midline. The second option, which would also reduce the dimension of the left cleft and the fistula, was chosen (Fig 6). A surgical splint was prepared accordingly.

A LeFort I osteotomy of lateral bone segments was performed under general anesthesia. Two lateral segments were mobilized. We tried not to devascularize

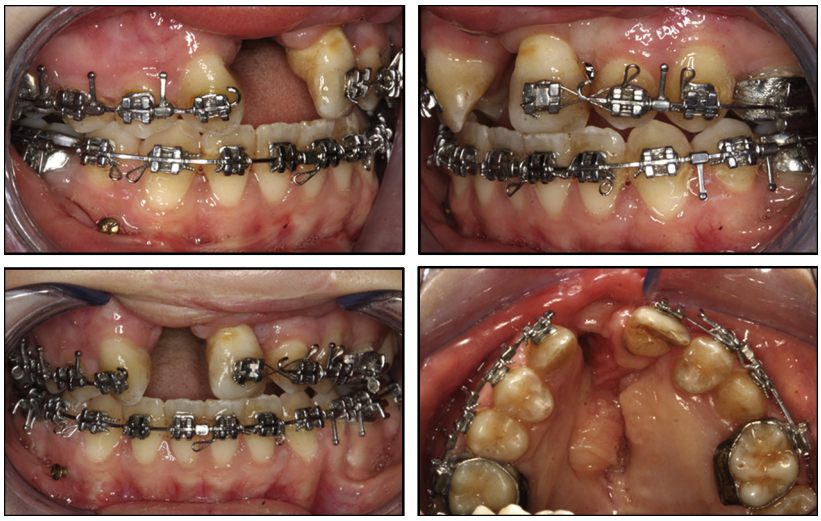

Fig 7. Postoperative intraoral photographs.

Fig 8. Intraoral photographs after orthodontic treatment.

the mobile premaxillary segment during the surgical dissection. Only the left intersegmental surfaces of the premaxilla and the left lateral segment were disepitelized so that after surgical movements the left fistula was repaired. The initial facial vertical height was recorded, and special attention was paid not to increase it. Because of the palatal scar, we experienced difficulty in positioning the left segment into the splint. Rigid and intermaxillary fixation was used for the posterior segments, whereas the premaxilla was fixed with intermaxillary fixation only. Intermaxillary fixation was discontinued after 2 weeks (Fig 7). During the finishing phase, intermaxillary elastics were used for refinement of the occlusion. In the last surgical intervention, the remaining right fistula was repaired with a buccal mucosal flap in conjunction with rhinoplasty and lip revision.

After orthodontic treatment, fixed retainers for both arches and a maxillary Hawley appliance were applied, and the patient was referred for prosthetic rehabilitation (Figs 8 and 9). The periodontal examination showed level discrepancies between the gingival margins of the maxillary right and left teeth. An esthetic crown lengthening procedure was planned to provide appropriate proportions of the anterior teeth, along with pleasing gingival symmetry. Gingival tissues of 2 mm from the gingival margins of the maxillary right canine and

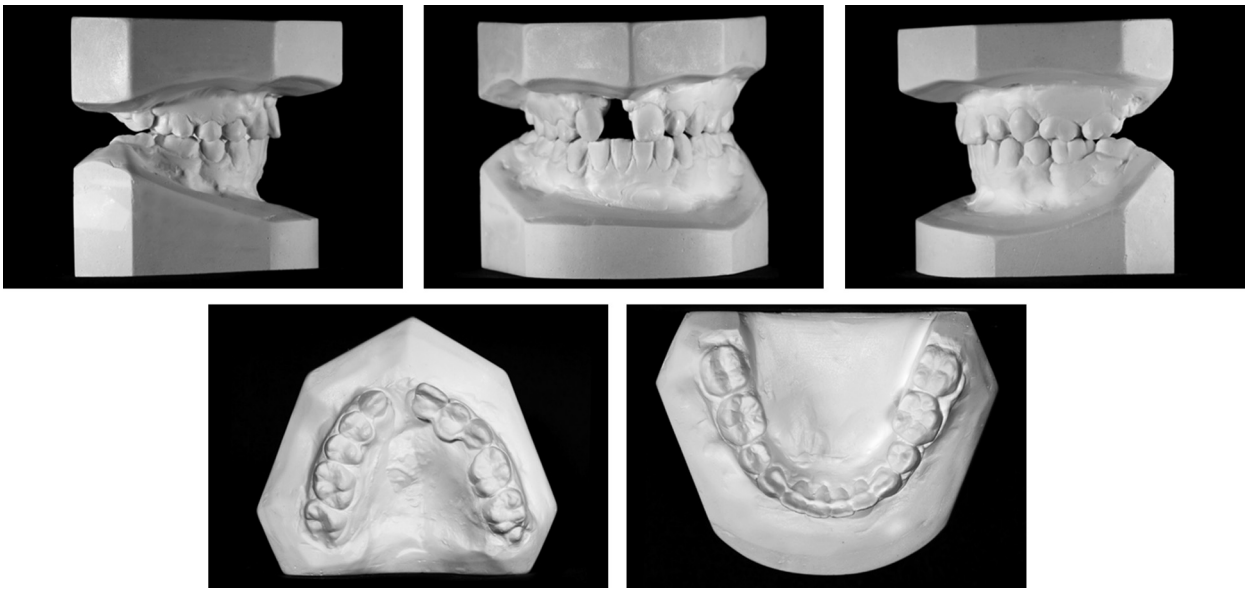

Fig 9. Posttreatment dental casts.

premolars were excised with an inverse bevel incision, in a scalloped pattern around the gingival margin. After 2 weeks of healing, the maxillary teeth (right first and second premolars and canine; left central incisor, canine, first premolar, and first molar) were prepared with a wide-chamfer finish line. After conventional clinical and laboratory procedures, a porcelain fused to metal fixed partial denture was fabricated and delivered to the patient for rehabilitation and stabilization of the maxillary dental arch.

TREATMENT RESULTS

At the end of interdisciplinary treatment, which lasted 4 years, the patient's frontal and lateral facial appearance was improved (Fig 10). Normal upper and lower lip relationships to the esthetic plane were achieved (UL-E plane, −3 mm; LL-E plane, −1 mm). The nasolabial angle decreased and approached the norm for white people (Fig 11, *B*; Table).

The maxillary constriction and posterior crossbite were mainly solved by orthodontic expansion, and the remaining transversal problem was solved by surgery. The intermolar and interfirst premolar distances increased from 38.3 to 49.7 mm and from 20.6 to 30.3 mm, respectively. The vertical deficiency of the lateral segments was corrected during orthognathic surgery, with both vertical translational and rotational movements of the segments. The left fistula was reduced by the movements of the premaxillary and left lateral segments toward each other, whereas the right fistula enlarged (Fig 10). However, at the end of treatment, both fistulae were closed, and the patient was satisfied with the result.

After orthodontic treatment, the maxillary and mandibular dental midlines were coincident with the facial midline. Final overjet and overbite were both 1 mm. A Class I molar relationship on the right side and a Class II molar relationship on the left side were achieved. The right canine was substituted for the lateral incisor, whereas the left first premolar was substituted for the left lateral incisor, because of the canine-premolar transposition. A fixed prosthesis served to rehabilitate the occlusion, achieve dental esthetics, and retain occlusal relationships.

The posttreatment panoramic radiograph showed good root parallelism (Fig 11, *A*). There was no evidence of root resorption. The posttreatment cephalometric evaluation showed that the lower facial height did not change. The maxillary incisor was proclined orthodontically. By the proclination of the premaxillary segment, the A-point moved palatally, and the SNA and ANB angles and the Wits appraisal decreased (Figs 11, *B*, and 12; Table).

The posttreatment follow-up of the patient after 1 year showed stable occlusal and facial results (Fig 13). No recurrence of the intraoral fistulae was detected.

DISCUSSION

Patients with cleft lip and palate have multiple functional and esthetic problems. Only a team approach can

Fig 10. Extraoral and intraoral photographs after prosthetic rehabilitation.

provide comprehensive treatment for them. A successful treatment requires not only interdisciplinary treatment planning, but also evaluation of the treatment progress; modifications of the treatment plan by the team members might be necessary because the patient's needs and priorities can change, or a step of the treatment might fail. Because our patient declined secondary bone grafting, the team decided not to procede with the objective of achieving a Class I molar relationship on the left side but to decrease the cleft and fistular dimensions at least on 1 side.

Two-dimensional treatment planning on conventional cephalometric radiographs is inadequate for the prediction of complex orthognathic treatments when both linear and rotational movements on different axes are performed together. Model surgery on articulators helps to simulate the movements of the dental arches in surgery; however, only the dentition is represented 3 dimensionally, and the skeletal component is missing. Because dental models do not depict the surrounding bones, the surgeon cannot visualize the effects of model position on the facial skeleton.[10] Advances in 3D imaging technology led to the development of new computerized tools for surgical planning.[6,9] Computer-aided 3D planning enables the craniofacial skeleton to be viewed when planning treatment, mobilizing osteotomized bone structures, visualizing interferences between bone segments, and predicting hard-tissue and soft-tissue changes.[11] Recent studies have proved the feasibility and accuracy of computer-aided 3D surgical simulations.[6,9] For our patient, the 3D virtual surgery on 3D computerized tomography allowed all team members to voice the pros and cons of each approach from their points of view. Therefore, it brought all team members into agreement with the preoperative surgical treatment plan and enhanced communication within the team. Furthermore, we were able to simulate different surgical alternatives and select the most appropriate plan, as mentioned by Gateno et al[9] as another

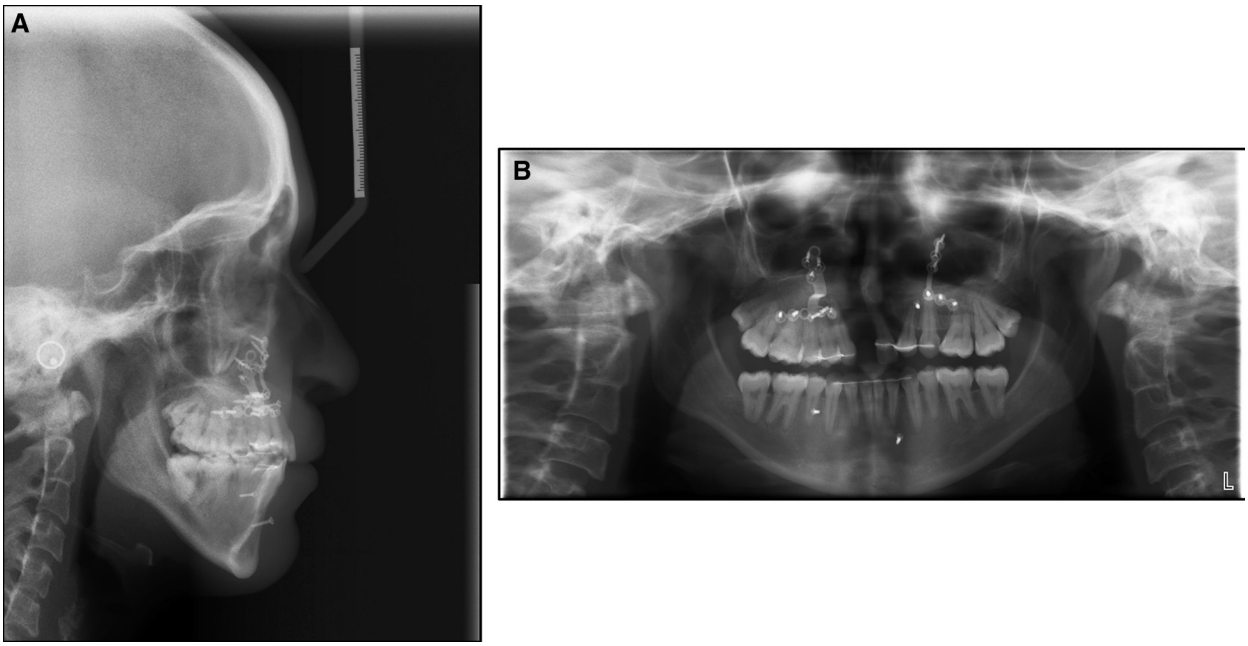

Fig 11. Posttreatment radiographs: **A,** lateral cephalometric radiograph; **B,** panoramic radiograph.

Table. Pretreatment and posttreatment cephalometric measurements

Cephalometric measurements	*Norm Mean ± SD*	*Pretreatment*	*Posttreatment*
SN-GoGn (°)	32.9 ± 5.2	44.9	44.8
SNA (°)	82 ± 3.5	82.9	79.3
SNB (°)	80.9 ± 3.4	77.5	76.4
ANB (°)	1.6 ± 1.5	5.4	2.8
Wits appraisal (mm)	−1 ± 1	7.9	3.5
U1-SN (°)	102.8 ± 5.5	84.2	102.9
IMPA (°)	95 ± 7	79.4	80.6
Overjet (mm)	2	−5	1
Overbite (mm)	2	1	1
UL-E plane (mm)	−6 ± 2	−5.5	−3.3
LL-E plane (mm)	−2 ± 2	2.1	−1.3

advantage of this method. Also, computer-aided surgical simulation enabled the surgeon to visualize the reality with which he had to deal in the operating room.

Maxillary constriction and posterior crossbite are common findings in patients with CLP. There are several options to correct these functional problems: slow or rapid maxillary expansion, surgically assisted orthodontic expansion, transpalatal distraction, and expansion during surgery.[12-14] Although our patient was an adult, we preferred to expand the maxillary arch with slow expansion because the integrity of the alveolar ridge could not be accomplished by secondary bone grafting. The main problem with orthodontic expansion is unwanted buccal tipping of the posterior teeth. Furthermore, occlusal plane changes can occur.[14] The patient also experienced buccal tipping of the teeth and maxillary segments caused by overexpansion; however, after removal of the quad-helix, a slight relapse occurred, and tooth inclinations were controlled during fixed orthodontic therapy before surgery.

Secondary grafting of the alveolar clefts enables restoration of the alveolar bone integrity, spontaneous eruption of the adjacent teeth to the cleft side, orthodontic tooth movement, or dental implant placement in the edentulous cleft area after grafting.[15-17] However, failure is also possible because of inadequate covering with the surrounding soft tissues, intensive scar formation, or large clefts. The success rate of autogenous bone grafting is controversial. Especially in adult patients where no tooth eruption will occur into the grafted bone, there is a high risk of resorption because of disuse atrophy.[14] According to Toscano et al,[18] the only factors involved in the stability of the graft seem to be dental age at the time of bone grafting and orthodontic therapy before and after grafting. This implies the importance of tooth movement to prevent postoperative bone resorption. Although bone grafting was proposed to this patient mainly to stabilize the maxillary segments, she declined this treatment. The benefit of alveolar grafting for this patient is open to dispute. We did not plan any tooth movement into the graft or placement of a dental implant, which would have preserved the bone, because she had multiple missing teeth, dental transposition,

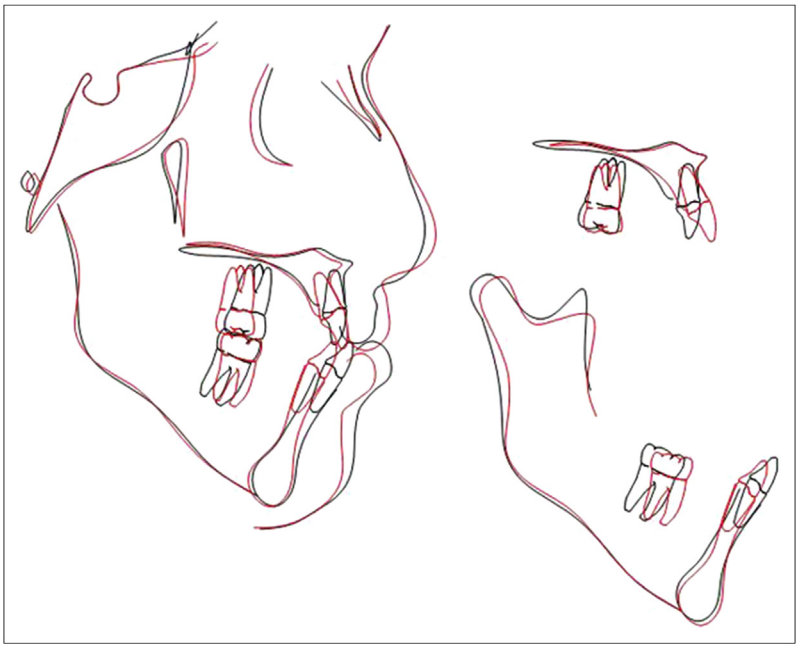

Fig 12. Pretreatment and posttreatment lateral cephalometric superimpositions.

and a hypomineralized incisor requiring extensive restoration to achieve functional and esthetic outcomes. Therefore, grafted bone might have been prone to resorption in the long term.

Segment distraction is an effective treatment alternative to reduce the width of wide alveolar clefts, especially before alveolar bone grafting.[19-23] This procedure is also indicated for patients who have undergone bone grafting resulting in survival failure or patients who do not desire iliac bone grafting.[21] During distraction, 3D vector control is important to achieve ideal results. It was reported that the distractors have insufficient transversal stability when applied unilaterally, with the potential risk of the lateral segment tilting palatally during the distraction.[20] Furthermore, the distraction vector can result in a flat and medially collapsed maxillary arch because of its curved architecture.[23] To prevent these side effects, expansion appliances, buccally extending metal bars, palatal arch bars, temporary anchorage devices, intraoral elastics, and specially designed archwise appliances can be used, or a 2-step distraction appliance addressing different vectoral problems can be applied.[20,21,23-27] However, some of these can be complicated procedures. Because the methods attempting to create an arch curvature during bone transport are relatively novel techniques, there are no randomized clinical trials showing their effectiveness on forming a curve, and the results are mainly based on case reports.

In our patient, the preoperative positions of both lateral segments necessitated sagittal, vertical, and transversal corrections. Pichelmayer and Zemann[24] described the risk of maxillary constriction during vertical distraction. Because of the difficulty in accurately controlling the transported segments in 3 dimensions and the possibility of a 2-step intervention, we did not want to reduce the cleft size with segmental distraction. Instead, we immediately positioned the left lateral segment anteriorly, combined with vertical and transverse movements after the segmental LeFort I osteotomy, to control translational and rotational movements of the segment and preserve dental arch curvature more readily than with a distraction procedure. This modified LeFort I osteotomy has been proposed to close the cleft gap, resolve oronasal fistulae, manage skeletal defects, and correct jaw deformities simultaneously in cleft patients.[28] However, the limiting effect of scar tissue on the lateral segment's movement should also be considered. Perhaps, for extensive movements of the maxillary segments, it would be better to choose distraction osteogenesis, because in the dentoalveolar region it allows lengthening of both bone and soft tissues. On the other hand, even though distracted, soft tissues can generate traction forces guiding the bone segment to an unfavorable position during the distraction period.[24]

Dental abnormalities are more frequent in patients with CLP than in the general population.[29] Our patient had 3 congenitally absent maxillary teeth accompanied by a canine-premolar transposition, which complicated

Fig 13. Extraoral and intraoral photographs 1 year after treatment.

the treatment and challenged the achievement of an esthetic and functional occlusal outcome. There is no consensus among dentists about the treatment of dental transpositions. Treatment is selected on an individual basis. Some authors suggest orthodontic correction of the transposition in the maxilla, whereas others support alignment of the teeth in their transposed positions.[30,31] For a partial transposition, correction of the transposition can be an option, whereas for a complete transposition, preservation of the initial position might be a better alternative. Because our patient had a complete transposition, we kept the teeth in their initial positions. It has also been reported that correcting a transposition can prolong the treatment time.[32]

Patients with CLP are likely to require fixed, removable, or combined prosthodontic rehabilitation to replace missing teeth, improve esthetics, or obturate the palatal deficiency. When the edentulous cleft site is not closed orthodontically or surgically, prosthetic treatment is required. Most cleft patients prefer some form of fixed prosthetic replacements. If a bone graft has been performed, implant-supported tooth replacement can be used. If the arch has not been stabilized with a bone graft and the teeth adjacent to the edentulous space have morphologic deficiencies, consideration can be given to fabricating a fixed partial denture.[33] In our patient, no palatal obturation was required, so a removable denture was not considered. Because of the many missing teeth, the left maxillary canine-premolar substitution, the malformed teeth, and the lack of grafting, fabrication of a fixed partial denture was considered. With a prosthesis, the missing teeth were substituted, the transpositioned teeth were reshaped, and stabilization of the maxillary dental arch was provided. At the 1-year follow-up, the fixed partial denture remained intact and functional, and no further complications were noted.

CONCLUSIONS

At the end of the interdisciplinary treatment, a functional occlusion, a harmonious profile, and patient

satisfaction were achieved. The results were stable 1 year after orthodontic treatment.

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