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CASE REPORT

Multidisciplinary Treatment of an Untreated Young Adult Patient with Unilateral Complete Cleft Lip and Palate

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ABSTRACT

We present the multidisciplinary treatment of a young adult patient with unilateral complete cleft lip and palate (UCLP). The patient with UCLP was 17 years old and had not applied for treatment before. He presented with a concave profile, lateral crossbite and a tete-a-tete overbite. After initial orthodontic treatment the patient's cleft lip and nose and afterwards his palate were operated on. During the orthodontic treatment the patient had a negative overjet of 6 mm, a residuel fistule in soft palate, maxillary and secondary nose base deficiency, also a severe alveolar cleft in the premaxilla. To fix these problems, the patient's maxilla was advanced by applying a Lefort-1 osteotomy, the secondary fistule in the soft palate was operated on and the alveolar defect was grafted with a biocollagen membrane, cansellous block graft and cansellous granular graft. The orthodontic treatment lasted 1 year following the orthognathic surgery. At the end of the orthodontic treatment Class I molar relationship was achieved on the right side and full Class II on the left side and also a 1 mm overjet and overbite. After a period of retention of 1.5 years some relapse occurred and delayed prosthetic treatment was performed by applying an adhesive bridge. Late term multidisciplinary treatment gave the UCLP patient a good appearance as well as psychological and social benefits.

Keywords: Untreated UCLP, adult UCLP, multidisciplinary treatment

INTRODUCTION

In developed countries most patients with cleft lip and palate undergo surgery early in life so the rare untreated cleft patients are mostly found in so-called third world countries. The problem of growth inhibition resulting from surgical treatment of the cleft lip and palate is a widely discussed topic.[1] Individuals with untreated cleft lip and palate shed light on how the untreated upper jaw develops. This case report presents the multidisciplinary management of an untreated unilateral complete cleft lip and palate in a young adult patient.

CASE REPORT

The patient was a 17-year-old boy with a residual 6.5 % of growth and development potential when we first met him working during the reconstruction of our faculty building. He presented an untreated unilateral complete cleft lip and palate. During the extraoral examination we observed a deviated nose to the right side of the face and a concave profile. Intraoral examination revealed an Angle Class III molar relationship on the right side and a Class II molar relationship on the left side with a tete-a-tete overbite and 2 mm of overjet. He exhibited a posterior cross-bite in the first premolar region on the left side and in the premolar and molar region on the right side. Upper left santral and lateral teeth were missing due to the alveolar cleft in the anterior region. There was an arch discrepancy of +11.5 mm in the upper arch and +3 mm in the lower arch (Figure 1, 2).

Lateral cephalometric analysis indicated a border Class 1 skeletal relationship (ANB: 0º) with excessive mandibular length (CoGn: 124 mm) and optimum maxillary length (Co-A: 91.5 mm). However, the maxilla was retrog-

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Figure 1. Intraoral and extraoral photographs of the patient at the beginning of the treatment (T1)

Figure 2. Panoramic radiograph of the patient at the beginning of the treatment (T1)

natic (FH-NA: 87.7º). Cephalometric measurements presented an optimal growth pattern (SNGoGn: 30.8º). The upper incisors and lower incisors were protrusive (U1-NA: 7.5 mm; L1-NB: 5,6 mm) (Figure 3, Table 1).

Treatment Plan and Procedure

In this case, the aim of the treatment plan was:

- Improvement of facial esthetics with the surgical repair of cleft nose, lip and palate,
- Advancement of the maxilla,
- Full fixed orthodontic treatment,
- Prosthodontic treatment of the missing teeth.

The surgical treatment approach for the patient was to operate on the cleft lip and palate with orthodontic alignment of the teeth. A modified Millard rotational technique for the lip was applied first. Then six months later a bi-layered closure of the hard and soft palate was performed using Von Langenbeck and modified

Figure 3. Lateral cephalometric radiograph of the patient at the beginning of the treatment (T1)

Figure 4. Intraoral and extraoral photographs of the patient before orthognathic surgery (T2)

Figure 5. Panoramic radiograph of the patient before orthognathic surgery (T2)

Table 1. Cephalometric and stone model measurements at the beginning (T1), before orthognathic surgery (T2), at the end of full fixed therapy (T3), after 1.5 years of retention period (T4)

Parameters	Norm values	T1	T2	T3	T4
SNA (º)	82.0	80.7	80.7	82.0	80.0
SNB (º)	80.0	80.7	83.0	82.5	82.0
ANB (º)	0.0	0.0	-2.3	-0.5	-2.0
Convexity (A-NPo) (mm)	2.0	-0.7	-4.1	-2.6	-2.6
Maxillary Depth (FH-NA) (º)	90.0	87.7	87.2	87.1	87.9
Co-A (mm)	92±2.7	91.5	92	95.5	92.5
Co-Gn (mm)	118±5.0	124	132	133.5	134.5
SN - GoGn (º)	32.0	30.8	26.6	27.5	29.5
Mandibular Body Length (Go-Gn)(mm)	94.4	83.1	93.8	94.4	96.4
U1 - NA (º)	22.0	34.9	20.0	28.5	26.3
U1 - NA (mm)	4.0	7.5	3.3	6.4	6.2
L1 - NB (º)	25.0	23.0	22.0	15.5	22.7
L1 - NB (mm)	4.0	5.6	5.0	4.3	5.6
Facial Convexity (G'-Sn-Po') (º)	12.0	13.5	-1.9	8.1	5.4
Upper Lip to E-Plane (mm)	-5.4	-3.6	-8.9	-6.5	-7.9
Lower Lip to E-Plane (mm)	-2.0	-1.7	2.3	0.7	-1.7
Nasolabial Angle (Col-Sn-UL) (º)	102.0	112.6	95.6	106.7	105.3
Interkanine width UW3 (mm)		29.0	31.0	32.0	32.0
Intermolar width UW6 (mm)		51.0	49.0	50.0	50.0

Figure 6. Lateral cephalometric radiograph of the patient before orthognathic surgery (T2)

Figure 7. Intraoral and extraoral photographs of the patient at the end of full fixed therapy (T3)

Figure 8. Panoramic radiograph of the patient at the end of full fixed therapy (T3)

Oxford techniques. For the protraction of the maxilla the patient was instructed to wear reverse headgear. However, advancement of the maxilla could not be achieved due to poor cooperation. It took 3 years of orthodontic treatment to align the maxillary and mandibular arches. However, a skeletal Class 3 relationship with negative overjet of 6 mm occurred. Furthermore, a lateral crossbite on the left side, severe alveolar cleft in the premaxilla, residuel fistule in the soft palate plus maxillary and secondary nose base deficiency were diagnosed (Figure 4-6). In order to fix these problems the patient underwent LeFort I osteotomy with advancement combined with alveolar grafting and soft tissue repair. The classic osteotomy lines above the apices of the teeth on both segments were achieved using rotating instruments and osteotomes to loosen the segments. A surgical splint was applied to the mandible and the loosened segments were placed over it in the new position

Figure 9. Lateral cephalometric radiograph of the patient at the end of full fixed therapy (T3)

Figure 10. Intraoral and extraoral photographs of the patient at the end of 1.5 years of retention period (T4)

Figure 11. Panoramic radiograph of the patient at the end of 1.5 years of retention period (T4)

Figure 12. Lateral cephalometric radiograph of the patient at the end of 1.5 years of retention period (T4)

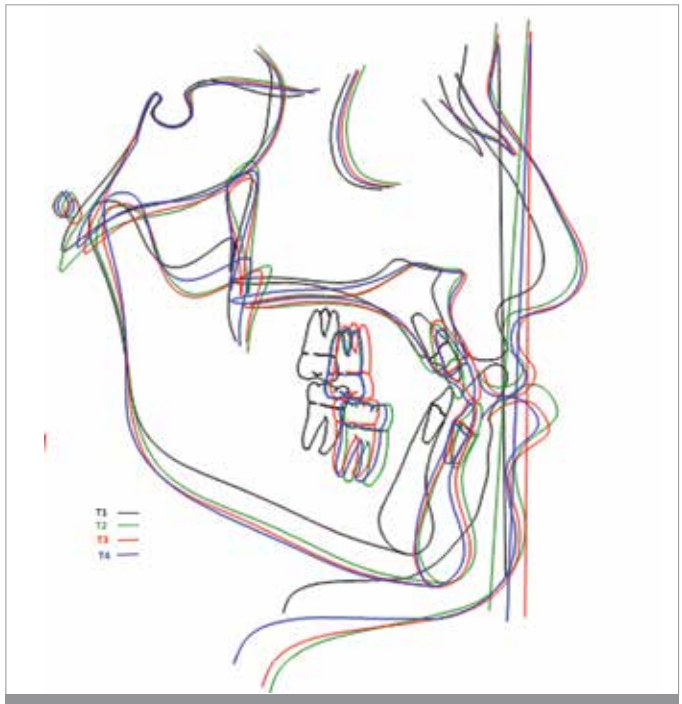

Figure 13. Total cephalometric superimpositions of the patient at the beginning (T1), before orthognathic surgery (T2), at the end of full fixed therapy (T3), after 1.5 years of retention period (T4)

using rigid miniplate fixation with mini screws. The cleft alveolus was reconstructed with a cancellous block graft (10 × 10 × 20 mm) and pre-hydrated collagenated heterologous cortico-cancellous chips (2–4 mm size of 4 cc Mp3 Tecnoss, Giaveno, Italy). A heterologous pericardium membrane (25 × 35 mm, Evoluation, Tecnoss, Giaveno, Italy) was used to cover the graft material. The soft tissues were closed with resorbable sutures. The intermaxillar fixation with elastics stayed for 6 weeks using a surgical splint. The healing was uneventful. After 1 year of orthognathic surgery, a Class I molar re-

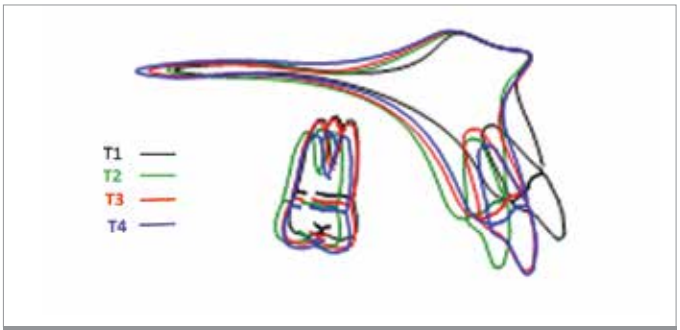

Figure 14. Local maxillary superimpositions of the patient at at the beginning (T1), before orthognathic surgery (T2), at the end of full fixed therapy (T3), after 1.5 years of retention period (T4)

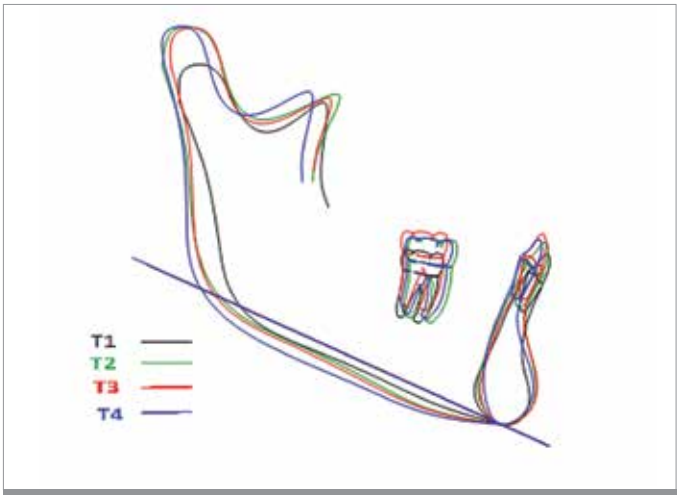

Figure 15. Local mandibular superimpositions of the patient at the beginning (T1), before orthognathic surgery (T2), at the end of full fixed therapy (T3), after 1.5 years of retention period (T4)

lationship was achieved on the right side, full Class II molar relationship on the left side as well as 1 mm of overjet and overbite (Figure 7–9). The prosthetic rehabilitation was delayed because the patient had to carry out his compulsory military service. The patient was instructed to use Hawley retainers full time as the retention protocol. The upper Hawley retainer also included acyrilic left central and lateral teeth to make up for the missing teeth in the cleft area.

After a retention period of 1.5 yearss some relapse occurred and the patient exhibited a tete-a-tete overbite, 0.5 mm of overjet and posterior crossbite on the left side. Intercanine and intermolar widths were stable both at the end of full fixed therapy and the retention period (Figure 10–12). The total and local superimpositions of the patient are shown in Figure 13–15. Delayed prosthetic treatment was performed with an adhesive bridge due to the patients' poor oral hygiene and financial resources, and this offered the usual characteristics in terms of functionality and esthetics. After the prosthetic treatment a full-covarage thermoplastic retainer was prepared for the maxillary arch to be used at night to preserve transversal stability.

DISCUSSION

One of the major problems in the treatment of cleft lip and palate is the disturbance of maxillary growth.[2] Several studies have been conducted on unoperated adult cleft lip and palate patients in order to determine if facial growth differences are due to intrinsic growth factors or surgical interventions, and there have been conflicting opinions.[3-12] The results of a study by Shetye and Evans[3] show differences in the measurements of the craniofacial structure between unoperated adult patients with complete unilateral cleft lip and palate and normal checks. Other studies indicate that some of degree of maxillary retrusion and arch contraction, as well as median facial dysplasia were seen in adult patients with CLP even without the interference of surgery.[6,11] However, the morphology of craniofacial structures in unoperated cleft patients is more favorable than that observed in surgically treated cleft patients indicating that disturbances of maxillary growth in surgically operated cleft patients were primarily due to surgical intervention.

In the literature review by Bardach and Salyer [12] it was concluded that undermining the soft tissues at the time of lip repair and exposing the bare bone at the time of palatal repair coupled with improper surgical execution and the resulting scar in the area of the denuded bone that is firmly attached to the palatal bone using Sharpey's fibers can all lead to retarded growth. In Mommaerts'[13] study it was stated that the type of surgery also affects the quantity of maxillary growth. Furthermore, Manna et al.[14] stated that numerous surgical procedures performed on the same patient can adversely affect the growth potential of the bone.

In our case, effective maxillary length was optimal even though the maxillary position was retrusive and there was a posterior crossbite in the first premolar region on the left side and in the premolar and molar region on the right side at the beginning of the treatment. In Shetye and Evans[3] study the size of maxilla was found to be normal and also somewhat prognathic in position in untreated adult cleft patients. Also Lambrecht et al.[1] concluded in their review article that the maxilla of patients with untreated clefts demonstrated a protruded position, probably caused by anterior tongue thrusts. The horizontal dimension was mildly reduced while the vertical dimension appeared normal in most cases.

In this particular case the surgical repair of the nose was insufficient and not symmetrical because of the redcued elasticity and severe deformity of the nasal cartilages. The only study[15] in literature comparing the results of primary cleft repair in infants with adults older than 17 years old reported that aggressive correction was possible in adults as maxillary growth was not a consideration while correction of the anterior part of the nasal deformity or septal deviation was more difficult than in infants. It was also stated that simultaneous palatoplasty should be chosen judiciously as it is more invasive and results in higher morbidity. Therefore, in our case palatoplasty was performed seperately 6 months after the lip and nose repair.

An oronasal fistule occurred after the cleft palate repair in this case. The clefts of adults are wider, and the surface of the hard palate is spiny, especially around the transverse palatine suture. In Morioka's study[15], it was reported that incidences of postoperative complications from simultaneous palatoplasty in adults were twice as high as that in the younger group. Ward and

James[16] also reported that the incidences of oronasal fistulas after palatoplasty were 38 percent in Sri Lanka.

In the present case after 3 years of orthodontic treatment including lip, nose and palate repair, 6 mm of negative overjet has occured and posterior crossbite has became more obvious, which could be related to residuel mandibular growth. Furthermore, the constriction effect of palatal surgery is one of the reasons for increased transversal discrepancy. During the repair of the palatal cleft, especially when using the Langenbeck technique, a large empty space is created between the elevated palatal mucosa and the denuded bone of the palatal shelves. The scar tissue that develops between these two structures contracts, and this can potentially result in transverse compression of the dentoalveolar dimensions. Ye et al.[10] stated that palatoplasty is the main cause of constriction of the maxillary arch while at the same time inhibiting the sagittal development of the anterior arch.

The present case underwent Lefort 1 osteotomy with advancement in order to improve the occlusion and the esthetic aspect. However, some relapse occured after a retention period of 1.5 years due to the contraction effect of the scar tissue. There was also a 2 mm increment of mandibular body length even though the patient was 21 years old at the end of full-fixed treatment.[17]

An adhesive bridge was used as prosthetic rehabilitation for the present case. Crowns and bridges supported by implants are the most popular forms of prosthetic restoration.[18-20] Much attention has been given recently to using implants[20,21] when rebuilding an edentulous alveolar cleft.[20,22] However, due to the necessity of bone augmentation as well as the long duration and the high cost of the procedure, implants were not appropriate for our patient.

Bridges in patients with cleft palates present a challenge with respect to hygiene because their broad surfaces can and do come into contact with the gingival tissues. Even if thoroughly veneered with porcelain there can stil be a build up of food and bacterial plaque, which can in turn cause the mucosa and periodontium to become infected.[19]

Given such limiting factors as the patient's poor oral hygiene[23] and his poor financial situation we opted for an adhesive bridge, which offered normal characteristics in terms of functionality and esthetics. Adhesive bridges are often described as being suitable for younger patients because they do not require significant preparation of the abutments and they can prove to be a viable alternative to implants in cases where there is a lack of alveolar bone.[24,25]

CONCLUSION

Late term multidisciplinary treatment provided the UCLP patient with a good appearance as well as psychological and social benefits.

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