UNIVERSIDADE DE SÃO PAULO FACULDADE DE ODONTOLOGIA DE BAURU

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Occlusal stability after orthodontic treatment in patients with UCLP: 3D analysis

Estabilidade oclusal após tratamento ortodôntico em pacientes com fissura labiopalatina unilateral: análise 3D

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Orientador: Prof. Dr. Prof. Dr. Marcos Roberto de Freitas

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FOLHA DE APROVAÇÃO

DEDICATÓRIA

Aos meus pais, Rosa Sánchez e Ramon Huanca,

Por me encorajar e apoiar durante este tempo longe de casa. Cada conquista na minha vida é de vocês também. Meu amor eterno, sempre no meu coração.

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Pelos conselhos e apoio na conquista dos meus sonhos. Por todo o carinho e companheirismo.

Aos meus colegas e amigos,

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"Seu trabalho vai preencher

uma parte grande da sua vida, e a única maneira de ficar realmente satisfeito é fazer o que você acredita ser um ótimo trabalho. E a única maneira de fazer um excelente trabalho é amar o que você faz."

-Steve Jobs

ABSTRACT

Occlusal stability after orthodontic treatment in patients with UCLP: 3D analysis

Objective: To evaluate the occlusal stability after orthodontic treatment in patients with unilateral cleft lip and palate (UCLP). Methods: The sample comprised 28 patients with non-syndromic UCLP (11 women, 17 men) orthodontically treated at the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo. Digital dental models were obtained before orthodontic treatment (T0, mean age 10.3 years), posttreatment (T1, mean age 18.7 years) and post-retention (T2, mean age 21.6 years). The following variables were measured at the three time-points using the Orthoanalyzer software: arch widths, arch length, arch perimeter, palatal depth, incisor irregularity index, overjet and overbite at cleft and non-cleft sides. Interphase changes were evaluated using ANOVA and Turkey tests (P<0.05). Results: In the maxillary arch, intercanine width, interpremolar width and palatal depth increased during treatment. In the mandibular arch, interpremolar width increased and arch perimeter and length decreased during treatment. Dimensional changes of maxillary and mandibular arches were stable after treatment. Maxillary and mandibular incisor irregularity decreased after treatment remaining stable in the long-term. During treatment, the overjet increased and the overbite decreased with an adequate stability after treatment. **Conclusion:** The occlusal changes of orthodontic treatment in patients with UCLP was stable approximately 3 years after debonding.

Key words: stability, relapse; orthodontics, corrective; cleft lip and cleft palate.

RESUMO

Estabilidade oclusal após tratamento ortodôntico em pacientes com fissura labiopalatina unilateral: análise 3D

Introdução: O conhecimento da estabilidade do tratamento ortodôntico é extremamente importante, e vastamente estudado na literatura. No entanto, são raros os estudos que avaliem a estabilidade da movimentação dentária em pacientes com fissura labiopalatina, e este estudo trará informações importantes. Objetivo: Avaliar a estabilidade oclusal após o tratamento ortodôntico completo em pacientes com fissura labiopalatina unilateral (UCLP). Material e Métodos: A amostra foi composta de 28 pacientes com UCLP não sindrômicos (11 mulheres, 17 homens) tratados com Ortodontia no Hospital de Reabilitação de Anomalias Craniofaciais da Universidade de São Paulo. Os modelos dentários digitais foram obtidos antes do tratamento ortodôntico (T0, idade média 10,3 anos), pós-tratamento (T1, idade média 18,7 anos) e pós-retenção (T2, idade média 21,6 anos). As seguintes variáveis foram medidas nos três pontos de tempo usando o software Orthoanalyzer: Largura do arco, comprimento do arco, perímetro do arco, profundidade palatina, índice de irregularidade dos incisivos, trespasse horizontal (overjet) e trespasse vertical (overbite) nos lados fissurado e não-fissurado. As mudanças interfase foram avaliadas utilizando os testes ANOVA e Turkey (P<0,05). Resultados: No arco maxilar, a largura intercaninos, a largura interpremolar e a profundidade palatina aumentaram durante o tratamento. No arco mandibular, a largura interpremolar aumento e o perímetro de arco diminuiu durante o tratamento. As mudanças dimensionais das arcadas maxilar e mandibular foram estáveis após o tratamento. A irregularidade dos incisivos maxilares e mandibulares diminuiu após o tratamento permanecendo estável a longo prazo. Durante o tratamento, o overjet aumentou e o overbite diminuiu com uma estabilidade adequada após o tratamento. Conclusão: As mudanças oclusais do tratamento ortodôntico completo em pacientes com UCLP foram estáveis aproximadamente 3anos após a remoção do aparelho.

Palavras-chaves: Estabilidade, Recidiva, Ortodontia, Fenda Labial, Fissura Palatina

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LIST OF ABBREVIATIONS AND ACRONYMS

- CPL Cleft Lip and Palate
- UCLP Unilateral Cleft Lip and Palate
- RME Rapid Maxillary Expansion

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1 INTRODUCTION

1 INTRODUCTION

Cleft lip and palate (CLP) is a congenital malformation that most frequently affects the face and oral. CLP is considered a relevant public health problem according to the World Health Organization. (Shaw, 2004) The prevalence of CLP is approximately 1 in 1000 live births, (WHO, 2002; Arosarena, 2007) (Trindade and Silva Filho, 2007) men have twice the number of cleft lip or cleft lip and palate than women.(Capelozza Filho and Silva Filho, 1992; Rego et al., 2003) The cleft lip and palate is a relatively common craniofacial malformation that occurs between the 4th and 12th weeks of intrauterine life, (Freitas et al., 2012a; Freitas et al., 2012b) and present a multifactorial etiology, by a combination of genetic factors (sex, race, heredity) and environmental factors (drugs, radiotherapy, hormones, nutritional conditions, infections). (Mossey, 1999; Freitas et al., 2012b) The treatment of patients with CLP should be initiated soon after birth and requires a team of specialists in an interdisciplinary approach. (Vlachos, 1996; Freitas et al., 2012a; Shaye et al., 2015) The primary plastic surgeries that are usually performed from 3 to 12 months of age, are cheiloplasty and palatoplasty; and the secondary alveolar bone graft performed between 9 and 12 years of age. In addition to primary plastic surgery, rehabilitation requires different specialties. The orthodontist follows the craniofacial growth treating complex malocclusions before and after alveolar bone grafting. (Freitas et al., 2012a)

The malocclusions observed in individuals with clefts present complexities that differentiate them from the occlusal irregularities of individuals without clefts. Besides the sagittal, transversal and vertical interarch discrepancies, patients with cleft lip and palate usually display tooth malpositions and dental anomalies.(de Lima Pedro et al., 2012; Rizell et al., 2020) In complete cleft lip and palate, the presence of a contraangulated and rotated maxillary central incisor is frequent, with the root apex mesially displaced to avoid the alveolar bone defect.(Freitas et al., 2012a) The cleft side maxillary canine also tends to present excessive mesial angulation determining a Class II sagittal relationship in the smaller segment, even in the presence of a Class III skeletal pattern. (Freitas et al., 2012a; Rizell et al., 2020) In unilateral complete cleft lip and palate, the maxillary complete cleft lip and palate. (Freitas et al., 2012a; Rizell et al., 2012a; Rizell et al., 2012a; Rizell et al., 2012a; Simões Holz et al., 2018) Maxillary dental crowding is frequently present due to sagittal and transverse maxillary deficiency. (Freitas et al., 2012a) Tooth agenesis and ectopic eruption are also frequently observed in individuals with clefts. (Lourenco Ribeiro et al., 2003; Letra et al., 2007; Menezes and Vieira, 2008) (Silva Filho et al., 1996) In permanent dentition, the agenesis of maxillary lateral incisor is the most common anomaly, followed by the agenesis of maxillary and mandibular second premolars. (Lourenco Ribeiro et al., 2003; Tortora et al., 2008; Dentino et al., 2012; Schwartz et al., 2017) Hypodontia of maxillary and mandibular second premolars is also common, with an increased prevalence compared to the general population. Ectopic eruption of maxillary first molar is seen in 20% of individuals with complete cleft lip and palate. (Silva Filho et al., 1996)

The planning for the comprehensive orthodontic treatment includes the decision between space closure of the region of missing maxillary lateral incisor by orthodontic mesialization of posterior teeth at the cleft side or space maintenance for implant placement. In cases of deviation of the maxillary midline and crowding in the greater segment may determine the need of asymmetric extractions in the maxilla.(Freitas et al., 2012a; Stonehouse-Smith et al., 2022) Orthodontic treatment for cleft patients is different from that of non-cleft patients, special management is needed and the stability of treatment should be considered.(Li, 2018) The dental arches should be coordinated during the comprehensive orthodontic treatment and adequate intercuspation should be achieved with positive overjet and overbite.(Freitas et al., 2012a)

Long-term occlusal stability in adult patients with UCLP have been investigated previously. Nicholson and Plint, studied relapse of RME and bone grafting; dental models were measured at start, end of RME and the follow-up models were taken at least one years after RME.(Nicholson and Plint, 1989) Ramstad et al, described the stability of orthodontic and prosthodontic in 22 patient treatment at six times with the final observation 13.5 years.(Ramstad and Jendal, 1997) Marcusson and Paulin, investigated posttreatment changes in patients with UCLP , who were divided into three groups according of type retention. (Marcusson and Paulin, 2004) Li, researched transverse changes dental arch and post retention stability in patients treated with quadhelix and edgewise appliance.(Li and Lin, 2007) Al-Gunaid investigate relapse tendency in patients with different type of maxillary arch form.(Al-Gunaid et al., 2008) Pucciarelli, compared stability of dental arches digitalized between patients with CLP and non-cleft.(Pucciarelli et al., 2020) Takahashi, evaluated the stability of treatment with at least 24 months after retention using 3D digitizer to measure both horizontal and vertical changes.(Takahashi et al., 2020) Recent study evaluated the occlusal outcome using the PAR index in patients with UCLP and bilateral cleft treated orthodontic treatment with/without orthognathic surgery.(Stonehouse-Smith et al., 2022) Nevertheless, little studies assess the stability of the maxillary and mandibular dental arch; few or no information available as to the relationship between stability of the incisor crowding index in patients with unilateral clefts lip and palate.

The stability in orthodontics is critical. Patients with cleft lip and palate have an aggravate scenario with soft tissue scars and fibrosis. Few studies assess the stability of orthodontic treatment in patients with clefts lip and palate. The objective of this study was to evaluate the occlusal stability 2 years after the comprehensive orthodontic treatment in patients with unilateral cleft lip and palate (UCLP). The hypothesis was that no changes in arch dimensions and tooth position are observed after debonding.

2 ARTICLE

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Occlusal stability after orthodontic treatment in patients with UCLP: 3D analysis

ABSTRACT

Objective: To evaluate the occlusal stability after orthodontic treatment in patients with unilateral cleft lip and palate (UCLP). Methods: The sample comprised 28 patients with non-syndromic UCLP (11 women, 17 men) orthodontically treated at the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo. Digital dental models were obtained before comprehensive orthodontic treatment (T0, mean age 10.3 years), posttreatment (T1, mean age 18.7 years) and post-retention (T2, mean age 21.6 years). The following variables were measured at the three time-points using the Orthoanalyzer software: arch widths, arch length, arch perimeter, palatal depth, incisor irregularity index, overjet and overbite at cleft and non-cleft sides. Interphase changes were evaluated using ANOVA and Turkey tests (P<0.05). Results: In the maxillary arch, intercanine width, interpremolar width and palatal depth increased during treatment. In the mandibular arch, interpremolar width increased and arch perimeter and length decreased during treatment. Dimensional changes of maxillary and mandibular arches were stable after treatment. Maxillary and mandibular incisor irregularity decreased after treatment remaining stable in the long-term. During treatment, the overjet increased and the overbite decreased with an adequate stability after treatment. **Conclusion:** The occlusal changes of orthodontic treatment in patients with UCLP was stable approximately 3 years after debonding.

Key words: stability, relapse; orthodontics, corrective; cleft lip and cleft palate.

INTRODUCTION

Cleft lip and palate (CLP) is a congenital malformation that most frequently affects the face and oral cavity.(WHO, 2002) CLP is considered a relevant public health problem according to the World Health Organization (Shaw, 2004) The prevalence of CLP is approximately 1 in 1000 live births.(WHO, 2002; Arosarena, 2007) The treatment of patients with CLP should be initiated soon after birth and requires a team of specialists in an interdisciplinary approach. (Vlachos, 1996; Freitas et al., 2012; Shaye et al., 2015) The orthodontist follows the craniofacial growth treating complex malocclusions before and after alveolar bone grafting. Besides the sagittal, transversal and vertical interarch discrepancies, patients with cleft lip and palate usually display tooth malpositions and dental anomalies. (de Lima Pedro et al., 2012; Rizell et al., 2020) In complete cleft lip and palate, the presence of a contraangulated and rotated maxillary central incisor is frequent, with the root apex mesially displaced to avoid the alveolar bone defect. (Freitas et al., 2012) The cleft side maxillary canine also tends to present excessive mesial angulation determining a Class II sagittal relationship in the smaller segment, even in the presence of a Class III skeletal pattern. (Freitas et al., 2012; Rizell et al., 2020) In unilateral complete cleft lip and palate, the maxillary midline is usually deviated to the cleft side. (Freitas et al., 2012; Simões Holz et al., 2018) Maxillary dental crowding is frequently present due to sagittal and transverse maxillary deficiency. (Freitas et al., 2012) Tooth agenesis and ectopic eruption are also frequently observed in individuals with clefts. (Lourenco Ribeiro et al., 2003; Letra et al., 2007; Menezes and Vieira, 2008) In permanent dentition, the agenesis of maxillary lateral incisor is the most common anomaly, followed by the agenesis of maxillary and mandibular second premolars. (Lourenco Ribeiro et al., 2003; Tortora et al., 2008; Dentino et al., 2012; Schwartz et al., 2017).

The planning for the comprehensive orthodontic treatment includes the decision between space closure of the region of missing maxillary lateral incisor by orthodontic mesialization of posterior teeth at the cleft side or space maintenance for implant placement. In cases of maxillary midline deviation associated with dental crowding in the greater segment, asymmetric extractions might be required. (Freitas et al., 2012; Stonehouse-Smith et al., 2022) Orthodontic retention for patients with orofacial clefts differs from non-cleft patients due the instability potential related to severe maxillary deficiency is often seen in patients with UCLP due to factors such constriction of scar and effect of surgeries.(Li, 2018) The dental arches should be coordinated during the comprehensive orthodontic treatment and adequate intercuspation should be achieved with positive overjet and overbite.(Freitas et al., 2012)

Long-term occlusal stability in adult patients with UCLP have been investigated previously.(Nicholson and Plint, 1989; Ramstad and Jendal, 1997; Marcusson and Paulin, 2004; Li and Lin, 2007; Al-Gunaid et al., 2008; Pucciarelli et al., 2020; Takahashi et al., 2020) Nicholson and Plint (Nicholson and Plint, 1989) compared the estability of RME with and without bone grafting. Dental models were measured before and after RME and at least one years after expansion. Transverse measurement of the maxillary arch had shown that bone grafting had not changed the overall pattern of relapse. The study by Ramstad and Jendal (Ramstad and Jendal, 1997) has evaluated the stability of orthodontic treatment 13.5 years after debonding and found that a reduction in the first molar width occurred during the initial five years after treatment. Marcusson and Paulin (Marcusson and Paulin, 2004) investigated posttreatment changes in patients with UCLP, reporting a larger reduction in the maxillary second premolar width. Li and Lin (Li and Lin, 2007) showed an increase in the maxillary arch width during treatment and a canine and premolar width relapse after retention. Al-Gunaid (Al-Gunaid et al., 2008) found that different types of maxillary arch shapes might play a stronger role in the stability after treatment. Pucciarelli et al. (Pucciarelli et al., 2020) compared the stability of dental arches between patients with CLP and non-cleft individuals, concluding that the maxillary dimensions were not stable one year after the orthodontic treatment. Takahashi (Takahashi et al., 2020) evaluated the stability of the orthodontic treatment at least 24 months after retention and found a horizontal relapse in the alveolar and dental regions at the second premolar region coupled with a vertical relapse on the cleft side of the central incisor, lateral incisor and canine.

The stability of orthodontic treatment is usually critical. Patients with cleft lip and palate have an aggravated scenario with soft tissue scars and fibrosis. The evidence on the stability of the orthodontic treatment in UCLP is scarce. Previous studies had mixed sample of cleft types and time length of retention. In addition, most of the previous studies has evaluated only the maxillary arch and the incisor irregularity index was not included. The objective of this study was to evaluate the occlusal stability after the orthodontic treatment in patients with unilateral cleft lip and palate (UCLP). The hypothesis was that no changes in arch dimensions and tooth position are observed after debonding.

MATERIAL AND METHODS

This observational study was approved by the institutional research committee Dental School, University of São Paulo of Bauru (process number 35403720.9.0000.5417) and Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru SP, Brazil (process number 35403720.9.3001.5441). Sample size calculation was based on an alpha of 5% and a beta of 20% to detect an average difference of 1mm with standard deviation of 1.18mm for the maxillary incisor irregularity index (Little Irregularity Index).(Freitas et al., 2017) The sample size calculation showed the need of 13 subjects.

Subjects with non-syndromic unilateral complete cleft lip and palate of both sexes were selected in a single center from 2019 to 2021. The inclusion criteria were: subjects who underwent orthodontic treatment, had dental models available and had at least the 2 years after debonding of orthodontic appliance, six months without using the retention. The exclusion criteria were orthognathic surgery and associated syndromes.

The final sample comprised 28 subjects (11 women, 17 men) with a mean age of 10.3 years before the comprehensive orthodontic treatment start. Lip repair was carried out from 3 to 8 months of life using Spina or Millard technique (Alberconi et al., 2018) and none had presurgical orthopedic treatment. The palatal repair was performed from 12 to 36 months of age using the Von Langenbeck technique. Alveolar bone graft using autogenous bone from the iliac crest was performed at a mean age of 12.2 ± 2.3 years.

Dental models were taken before orthodontic treatment (T0), after treatment (T1) and 2.86 \pm 1.5 years after debonding (T2). The mean age at T0, T1 and T2 were: 10.3 \pm 3.3, 18.7 \pm 2.8 and 21.6 \pm 2.6 respectively.

Non-surgical orthodontic treatment was performed in all patients at a single center. Orthodontic treatment was performed in two phases. Before the alveolar bone graft procedure, rapid maxillary expansion (RME) was performed in all patients. All patients underwent the same activation protocol of 1 complete turn a day (2/4 screw activation in the morning and 2/4 screw activation in the evening) for 7 days.

Nonsurgical compensatory orthodontic treatment started 4 months after bone graft procedure using preadjusted multibracket orthodontic appliances. Roth prescription with 0.022×0.025-inch slot brackets was used. The sequence of level and alignment was 0.014 and 0.016 NiTi arch wires, 0.016 and 0.018 and 0.019×0.025-inch stainless-steel wires. Hawley retainers were prescribed in the maxillary arch and canine-to-canine fixed retainer were bonded in the mandibular arch. All patients were instructed to wear Hawley retainer full-time for 12 months and during night-time for another 12 months. The mandibular retainer was instructed to be maintained at least for 2 years. Posttreatment consultations were performed every 6 months within 2 years.

Dental models were obtained before the orthodontic treatment (T0), after the orthodontic treatment (T1) and at least 2 years after debonding (T2). All dental models were digitized using an R700 3-dimensional scanner (3 Shape, Copenhagen, Denmark). The imagens were saved in STL format and measurements were performed using the 3Shape OrthoAnalyzer 3-dimensional software.(Fig. 1, 2 and 3) The following variables were measured in both the maxillary and mandibular dental models: intercanine, interpremolar, and intermolar widths (Fig 4-A), arch perimeter (Fig 4-B), arch length (Fig 4-C), palatal depth (Fig 4-D), Little irregularity index (Fig 5-A), the overjet and the overbite at the cleft and non-cleft sides (Fig 5-B).

For the error study, one operator (J.H.S.) performed all measurements and 30% of the sample was measured twice with an interval of at least 1 month. The intra-rater reliability was assessed using intraclass correlation coefficients (ICC).(Shrout and Fleiss, 1979) The Dahlberg formula was used for evaluating the casual error.

Statistical analyses

Means and standard deviations were calculated for all measurements at T0, T1 and T2. Normal distribution of all variables was verified using Kolmogorov-Smirnov tests. Interphase changes were evaluated using Analysis of variance (ANOVA), followed by Turkey tests. (Pandis, 2015) The statistical analyses were performed using Statistica software (version 10.0, StatSoft, Tulsa, Okla, EUA), considering a significance level of 5%.

RESULTS

Table I shows the results of the error study. All variables had excellent intrarater agreement with ICC varying from 0.997 to 1. The casual error varied from 0.06 (overbite non-cleft) to 0.41 (palatal depth).

Significant increases from T0 to T1 were found to maxillary intercanine width, maxillary interpremolar width, mandibular interpremolar width, palatal depth and overjet at both cleft and non-cleft sides. On the other hand, mandibular arch perimeter, mandibular arch length, incisor crowding and overbite at the non-cleft side decreased during treatment (T0-T1).

All variables remained stable from T1 to T2.

DISCUSSION

Digital dental models were used in this study permitting to perform precise linear measurement including the palatal depth. Studies comparing measurements in digital dental models and conventional plaster models have concluded that three-dimensional images are clinically acceptable and reproducible. (Goonewardene et al., 2008; Leifert et al., 2009; Bootvong et al., 2010) The error study showed an adequate intra-examiner reproducibility. This study presented difficulties to collecting longitudinal data. Few patients return for orthodontic checkups after finishing the use of retainers, thus there were few post-retention dental models. Prosthodontic section provided support in finding the sample once many patients return many years after orthodontic treatment to accomplish a prosthesis.

То orthodontic prevent relapse, retention is required after the treatment.(Littlewood et al., 2016) The minimal retention time recommended is 2 years. (Lang et al., 2002) In our sample, all patients were instructed to use Hawley retainer in the maxillary arch and fixed bonded retainer for mandibular arch for at least 2 years after debonding. The average orthodontic treatment time, including pre and post-bone graft intervention, was 8.46 years. This treatment time was extensive but similar to a previous study showing an orthodontic treatment time ranging from 3.3 to 8.5 years. (Semb et al., 2005) The mean follow-up time between T1 and T2 was 2.86 years. The limitation of this study is that 17 out of 28 patients still had the 3x3 bar bonded in the mandibular arch. However, the stability in the maxillary arch is more

critical than in the mandibular arch in subjects with UCLP. In the maxillary arch, the follow-up dental models were taken at least 6 months after the interruption of the retention.

The post-retention relapse in our study was smaller compared to previous studies (Nicholson and Plint, 1989; Ramstad and Jendal, 1997; Marcusson and Paulin, 2004; Al-Gunaid et al., 2008) These differences could be explained by different post-treatment observational time and different retention protocols. In addition, in our study the retention was controlled every 6 months stimulating the cooperation in wearing the retainers. Nicholson and Plint (Nicholson and Plint, 1989) reported maxillary width relapses, although patients were treated only with rapid maxillary expansion and had no fixed appliance therapy. The cases without posterior retention showed a severe degree of relapse even after bone grafting. (Nicholson and Plint, 1989) In other study, patients used fixed prothesis as a retention of orthodontic treatment and reported maxillary width relapses. (Ramstad and Jendal, 1997) The scar tissues in the palate, the pressure from the facial muscles and the traumatic effect of the surgical correction of the cleft palate have a stronger influence on the transverse growth of the maxilla compared to removable orthodontic retentions.(Marcusson and Paulin, 2004)

During treatment, the maxillary arch width significantly increased in the canine and first premolar regions (Table II). Maxillary arch expansion is the most common method for relieving crowding and transversal inter-arch relationship. In the current study, although the dental arch width decreased after treatment, the magnitudes were smaller than previous studies. The dental arch width decreased less than 1 mm after debonding and these changes were not statistically significant. The transversal changes remained stable after treatment. These findings might have occurred because the expansion was adequate and the retention protocol was properly used. Factors such as overexpansion and an adequate occlusion achieved by comprehensive orthodontic treatment might have influenced the stability in the maxillary arch.(Li and Lin, 2007) Our results are in agreement with a previous study showing a significant increase in the canine and premolar width during treatment and no significant changes after retention in individuals with CLP. (Takahashi et al., 2020) Ramstad and Jendal (Ramstad and Jendal, 1997) found that most of the posttreatment relapse occurred within the initial five years and were greater in the first molars followed by premolar and canines regions.

The palatal depth increased during the treatment, reflecting posterior teeth extrusion during the treatment time. Previous studies reported that the palate is shallower in individuals with cleft lip and palate compared to non-cleft individuals. (Ayub et al., 2016; Takahashi et al., 2020) Palatal depth decreased immediately after expansion in UCLP (Ayub et al., 2016). However, our study included a longer time of comprehensive orthodontic treatment after RME and bone grafting. The palatal depth remained stable after treatment. A previous study revealed no significant changes in the palatal height during and after orthodontic treatment in patients with oral clefts. (Takahashi et al., 2020)

The mandibular interpremolar width increased during treatment. Patients presented moderate dental crowding in the mandibular arch explaining some degree of traversal expansion that was observed during treatment. The transversal changes remained stable after treatment. These results are in agreement with the study by Li and Lin (Li and Lin, 2007). Mandibular arch perimeter and length decreased significantly during treatment. The changes remained stable after debonding. No previous studies on the mandibular arch perimeter and length in patients with oral clefts were found. In studies with non-cleft patients the mandibular arch perimeter and length decreased.

The severe maxillary and moderate mandibular incisor crowding index decreased during treatment. These changes were maintained after treatment in both arches and might be related to the proper use of orthodontic retainers. The retention protocol was similar with previous studies (Li and Lin, 2007; Al-Gunaid,2008). No previous studies on the stability of incisor crowding in patients with oral clefts were found. In studies with non-cleft patients the incisor crowding and relapse is usually greater in the mandibular arch. Differently, in our study the incisor crowding was greater in the maxillary arch.

The overbite at the non-cleft side slightly decreased during treatment showing aposttreatment stability. A previous study reported that the overbite remained stable at 5.6 years follow-up, agreeing with our results. (Marcusson and Paulin, 2004) Overjet increased significantly during treatment at both cleft and non-cleft sides. Some patients had a negative overjet before treatment. Changes remained stable after treatment. Our outcomes are in disagreement with a previous study (Marcusson and Paulin, 2004) reporting an overjet decrease after debonding in patients with orofacial clefts. These differences are probably due to age differences considering patients were at a younger age compared to ours and deffcient maxillary growth could have influenced the posttreatment relapse.

Further studies including longer post-treatment time after the retention protocol are recommended. The influence of debonding age on the stability-relapse in UCLP should also be investigated.

CONCLUSION

The occlusal stability of orthodontic treatment in patients with UCLP remained stable approximately 3 years after debonding. The correction of maxillary incisor malposition showed an adequate stability 6 months after the retention interruption.

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Figure captions

Fig. 1 Frontal images of the digital models of 3 subjects showing changes from T0 to T2.

Fig. 2 Upper occlusal images of the digital models of 3 subjects showing the changes from T0 to T2.

Fig. 3 Lower occlusal images of digital models of 3 subjects showing changes from T0 to T2.

Fig. 4 - Measurement of arch size: **A**, arch widths were intercanine, inter-premolar and inter-first molar width. The intercanine width was measured between the cup tips of canines or estimated cusp tips in cases of wear facets. The inter-premolar width was measured between buccal cusp tips of first premolars ; the inter-first molar width was measured between mesiobuccal cusp tips of first molars; **B**, arch perimeter was the sum of the 4 segments from mesial aspect of the right permanent first molar to the mesial aspect of the contralateral tooth; **C**, arch length (green arrow) was measured on the horizontal plane from mesial aspect of the permanent first molars to a point between the central incisors; **D**, palatal depth (red arow) was measured from a line passing through the mesial gingival papilla of the permanent first molars to the deepest point on the palate, perpendicular to arch length.

Fig. 5 A, Little's irregularity index was measured for both arches according to the methods of Little et al.; **B**, overbite and overjet on the cleft and non-cleft sides, were measured on a slice passing through the center of the maxillary right and left central incisors.



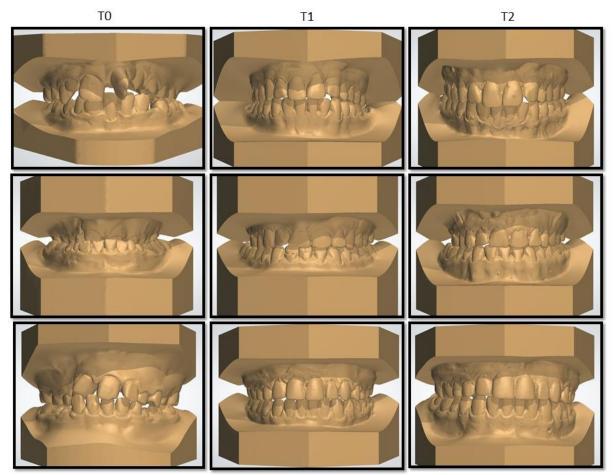
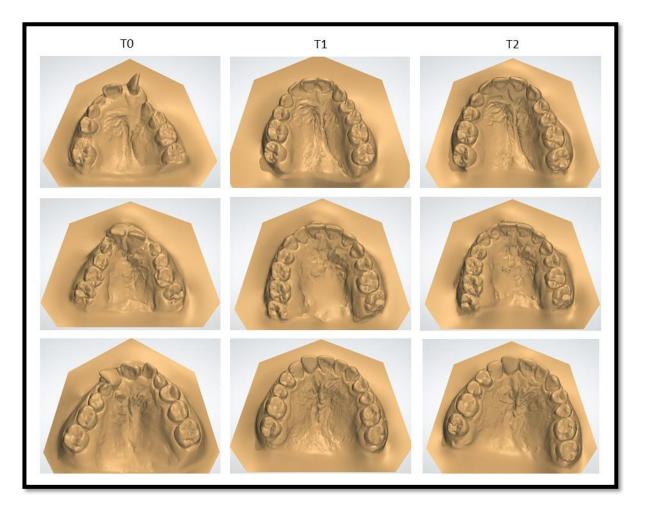
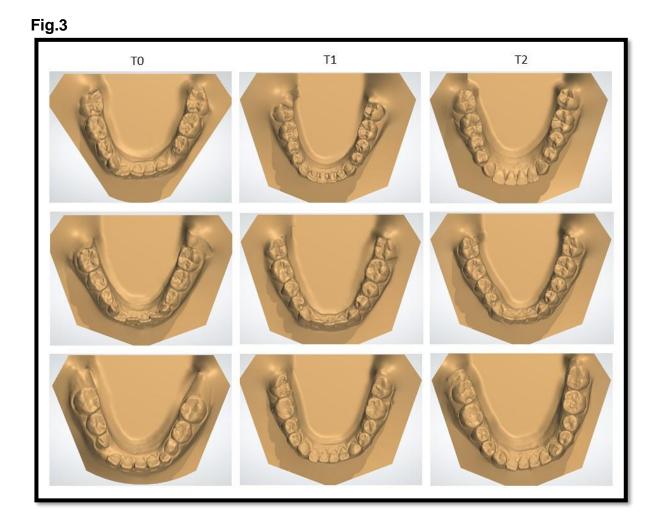
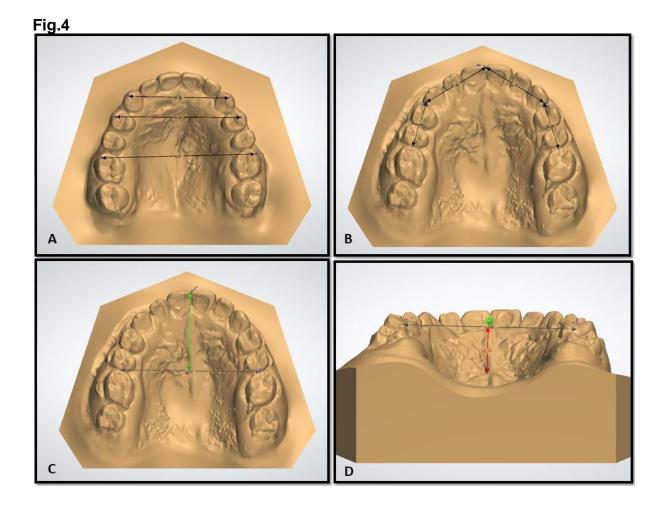


Fig.2







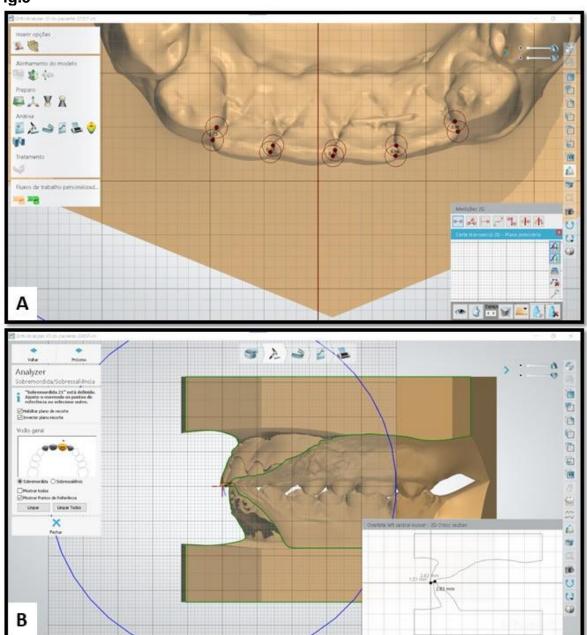


Fig.5

Variable	Measurement 1		Measurement 2		ICC	Dahlberg	Ρ			
	Mean	(SD)	Mean	(SD)						
Arch dimensions										
Maxilla										
3-3 width	30.86	3.58	30.83	3.57	1.000	0.08	0.130			
4-4 width	41.27	3.60	41.22	3.59	0.999	0.11	0.086			
6-6 width	51.77	3.14	51.70	3.14	0.999	0.09	0.003			
Arch perimeter	68.85	6.57	68.84	6.63	0.999	0.21	0.858			
Arch length	23.29	3.22	23.21	3.20	0.998	0.13	0.013			
Palatal depth	14.91	2.86	14.74	2.63	0.977	0.41	0.130			
Little	7.52	8.06	7.52	8.05	1.000	0.12	0.974			
Mandible										
3-3 width	27.47	1.73	27.43	1.72	0.998	0.07	0.028			
4-4 width	35.63	2.46	35.48	2.40	0.994	0.19	0.002			
6-6 width	46.61	2.18	46.53	2.17	0.996	0.14	0.040			
Arch perimeter	65.00	7.51	64.93	7.57	1.000	0.12	0.044			
Arch length	22.25	3.94	22.17	3.94	0.999	0.10	0.002			
Little	3.94	2.27	3.96	2.24	0.997	0.13	0.685			
Anterior relationship										
Overbite cleft	1.12	1.76	1.11	1.76	0.999	0.07	0.512			
Overbite	1.90	1.61	1.90	1.62	0.998	0.06	0.881			
Overjet cleft	0.79	3.11	0.85	3.09	0.997	0.16	0.151			
Overjet	1.60	2.69	1.64	2.72	0.997	0.14	0.248			

Table I. Error study for all variables assessed with intraclass correlation coefficients (ICC)

3-3, Intercanine; 4-4, interfirst premolar; and 6-6, intermolar

Variables	Т0	T1	T2	Р				
Variables	Mean (SD)	Mean (SD)	Mean (SD)					
Maxilla								
3-3 width	28.07 (2.89) ^A	32.51 (3.97) ^B	32.19 (3.78) ^B	0.000*				
4-4 width	38.60 (3.58) ^A	42.95 (3.01) ^B	42.38 (2.88) ^B	0.000*				
6-6 width	51.34 (4.09)	53.33 (3.55)	53.03 (3.41)	0.109				
Arch perimeter	69.19 (5.95)	70.67 (6.99)	70.26 (6.46)	0.106				
Arch length	23.37 (3.19)	23.87 (3.53)	23.75 (3.20)	0.498				
Palatal depth	12.74 (2.60) ^A	14.60 (3.35) ^B	14.85 (3.45) ^B	0.003*				
Little	13.97 (7.34) ^A	2.70 (1.66) ^B	3.37 (1.49) ^B	0.000*				
Mandible								
3-3 width	26.32 (2.38)	26.81 (2.26)	26.73 (2.10)	0.682				
4-4 width	33.56 (2.77) ^A	36.46 (1.99) ^B	36.11 (1.89) ^B	0.000*				
6-6 width	46.19 (2.85)	46.90 (2.95)	46.93 (3.03)	0.652				
Arch perimeter	67.44 (6.00) ^A	64.11 (6.95) ^B	63.65 (6.97) ^B	0.002*				
Arch length	23.52 (3.05) ^A	21.39 (3.63) ^B	21.24 (3.45) ^B	0.003*				
Little	5.94 (2.22) ^A	2.49 (1.66) ^B	3.33 (1.61) ^B	0.000*				
Anterior relationship								
Overbite cleft	1.64 (2.53)	1.29 (0.75)	1.37 (0.74)	0.688				
Overbite	2.97 (2.07) ^A	1.26 (0.74) ^B	1.59 (0.83) ^B	0.000*				
Overjet cleft	-1.04 (3.40) ^A	2.75 (0.91) ^B	2.61 (1.10) ^B	0.000*				
Overjet	0.54 (3.39) ^A	3.17 (0.91) ^B	2.91 (1.12) ^B	0.000*				

Table II. Comparison of the studied variables between the three stages (repeated-measures ANOVA, followed by Tukey tests).

Different letters in the same row indicate the presence of a statistically significant difference between the stages.

3-3, Intercanine; 4-4, interfirst premolar; and 6-6, intermolar.

* Statistically significant for p<0.05.

3 DISCUSSION

3 DISCUSSION

For the success of treatment, it is important to obtain primarily dental casts capable of providing prior information for the case study, and to be part of the patient's documentation. The dental casts demonstrate the inter-arch relationship in the sagittal, transverse and vertical directions, as well as intra-arch irregularities such as malposition and crowding. The evaluation of plaster models is an important method for inter-center studies of treatment results. (Molsted et al., 2005; Bartzela et al., 2010) The study on patients with cleft has been carried out using a plaster model and imaging exams, such as photographs and radiographs for dimensional anatomical measurements. Plaster casts are associated with the inconvenience of storing and transporting documentation.(Rosati et al., 2012)

Digital dental models were used in this study permitting to perform precise linear measurement including the palatal depth. Studies comparing measurements in digital dental models and conventional plaster models have concluded that threedimensional images are clinically acceptable and reproducible. (Goonewardene et al., 2008; Leifert et al., 2009; Bootvong et al., 2010) The error study showed an adequate intra-examiner reproducibility.

То prevent relapse, retention is required after the orthodontic treatment.(Littlewood et al., 2016) The minimal retention time recommended is 2 years. (Lang et al., 2002) In our sample, all patients were instructed to use Hawley retainer in the maxillary arch and fixed bonded retainer for mandibular arch for at least 2 years debonding. The average orthodontic treatment time, including pre and postbone graft intervention, was 8.46 years. This treatment time was extensive but similar to a previous study showing an orthodontic treatment time ranging from 3.3 to 8.5 years.(Semb et al., 2005) The mean follow-up time between T1 and T2 was 2.86 years. The limitation of this study is that 17 out of 28 patients still had the 3x3 bar bonded in the mandibular arch. However, the stability in the maxillary arch is more critical than in the mandibular arch in subjects with UCLP. In the maxillary arch, the follow-up dental models were taken at least 6 months after the interruption of the retention.

The post-retention relapse in our study was less than to other researchers (Nicholson and Plint, 1989; Ramstad and Jendal, 1997; Marcusson and Paulin, 2004; Al-Gunaid et al., 2008) This could be due to the short post-treatment time in this research, the prescribed retention protocol, the patients' cooperation in wearing retainers and the retention control every 6 months in our department. On another side, Nicholson (Nicholson and Plint, 1989) reported dental width relapsed, however, patients were treated only with rapid maxillary expansion and had no fixed appliance. The cases without posterior retention showed a severe to almost total degree of relapse, that some form of retention is far more important in maintaining expansion of maxillary arch than bone grafting. (Nicholson and Plint, 1989) In other research, the patients used only fixed prothesis as retention. (Ramstad and Jendal, 1997) Other study in which patients used different types of retention or without retention. (Marcusson and Paulin, 2004) However, previous study reported the scar tissues in the palate, pressure from the facial muscles, and the traumatic effect of the surgical correction of the cleft palate have the stronger influence on the transverse growth of the whole maxilla that the type of retention after orthodontic treatment. (Marcusson and Paulin, 2004)

During treatment, the maxillary arch width significantly increased, in the canine and first premolar regions (Table II). Maxillary arch expansion is the most commonly used method for relieving crowding and transversal inter-arch relationship. Current study, the dental arch width of each region decreased in post-treatment, the magnitudes were smaller than previous research. The dental arch width decreased less than1 mm post-retention, which was not statistically significant. The transversal changes remained stable after treatment. This may be because the expansion was adequate and retention protocol was used properly. Factors such as overexpansion and good buccal occlusion achieved by full fixed appliances after orthodontic treatment decrease the relapse tendency of upper arch. (Li and Lin, 2007) Takahashi (Takahashi et al., 2020) showed a significant increase canine and premolar width during treatment, no changes after retention in the CLP group, consistent with our findings. Ramstad and Jendal, (Ramstad and Jendal, 1997) found that most of the post-treatment change had taken place during the initial five years, to be greatest in first molar followed premolar and canines region. This would be in discordance with our study which found no change in molar region and found stability in dental widths

after 2 years debonding. Li (Li and Lin, 2007) reported canine and premolar width increase during treatment and decreased after retention, disagreement with our study.. Al-Gunaid,(Al-Gunaid et al., 2008) reported for group A (symmetrical arch), no change in treatment and no retention relapse for canine width. For group B increase for canine and premolar width, then relapse for canine and stability for premolar. For group C (collapse arch) a large increase in premolars and relapse in post- retention. The outcome of relapse which may be due to the fact that B and C groups presented collapse of segments in the upper arch. Disagree because the treatment changes remain stable in the post-retention period in our research. Disagreed with study of Pucciarelli (Pucciarelli et al., 2020) showed statistically significant relapse in the intercanine measument. Concluded that the maxillary dimensions were not stabilized after 1 year of orthodontic and prosthodontic treatment in UCLP group. The cleft area is the most vulnerable to orthodontic relapse.

The palatal depth increased during the treatment, reflecting some orthopedic effects. Studies previous has been reported that the palate in cleft lip and palate patients is shallower than in non-cleft. (Ayub et al., 2016; Takahashi et al., 2020) Palatal depth decreased after expansion in children with cleft, (Ayub et al., 2016) this differs from our study, which showed an increase after treatment. Few studies have involved longitudinal investigations. The post-retention outcome in our study was stable. Different from Takahashi,(Takahashi et al., 2020) revealed no changes in the palatal height during treatment and after least 24 months after retention were used.

The mandibular inter premolar width increased during treatment, the increase was significant. This study, patients presented moderate lower arch crowding, there was a need for lower arch widening in patients with UCLP. The transversal changes remained stable pos-retention. Li (Li and Lin, 2007) reported inter-premolar width increase during treatment and was stable post-retention results, agreed with our study. Mandibular arch perimeter and length decreased; the decrease was significant. The changes remained stable pos-retention. Maintenance of lower interdental width during orthodontic treatment is considered important to maintain treatment stability. Lower arch expansion is rarely necessary during orthodontic treatment in patients with UCLP.(Li and Lin, 2007) However, correction of dental malposition and good occlusion may induce changes in these measurements.

The incisor crowding index decreased, the decrease was considerable and statistically significant in the maxillary arch. The crowding was severe in maxillary arch. Mandibular incisor crowding index decreased. The decrease was considerable and statistically significant in the mandibular arch because arch had moderate crowding before treatment. The incisor crowding index in both dental arches decreased after treatment, mandibular crowding was smaller than maxillary. The changes were maintained in post-retention in both arches, this may be due to the fact that patients used the retainers properly. The retention protocol was similar those other studies (Li and Lin, 2007; Al-Gunaid,2008). However, these studies did not analyze this variable. No previous studies in in patients with cleft were found that reported about incisor crowding index. In studies with patients non-cleft,(Massaro et al., 2018) the incisor crowding was greater in the mandibular arch. Different from our study, which was greater in maxillary arch. Previous studies with patients non-cleft,(Massaro et al., 2018) reported increases of anterior crowding in longitudinal follow-ups of subjects with normal occlusion.

Overbite non-cleft decreased after treatment. Some patients had a deep bite at the beginning of treatment. At pos-retention increased slightly, but not significantly. Marcusson,(Marcusson and Paulin, 2004) reported that the overbite remained stable at 5.6 years follow-up, agree with this. Overjet increased significantly after treatment on both cleft and non-cleft. Some patients had a negative overjet at the beginning of treatment. Changes remained stable pos-retention. Marcusson,(Marcusson and Paulin, 2004) reported relapse, that overjet decreased, disagree with the result of this variable.

Few studies have been reported the stability of both dental arches and crowding index after orthodontic treatment in patients with repaired UCLP. Further studies including longer post-treatment time after the retention protocol are recommended.

CONCLUSION

4 CONCLUSION

The occlusal stability of orthodontic treatment in patients with UCLP remained stable 2.8 years after debonding. The correction of maxillary incisor malposition showed an adequate stability 6 months after the retention interruption.

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