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CASE REPORT

Tooth retention management in a patient with cleidocranial dysplasia

Manejo de retenciones en paciente con displasia cleidocraneal

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ABSTRACT

Cleidocranial dysplasia, a disease that disrupts bone growth and therefore limits the resorption of deciduous teeth, results in the non-eruption of the permanent dentition and interferes with facial aesthetics and functions. The following case describes a female patient with cleidocranial dysplasia treated at the Department of Orthodontics of Mexico Children's Hospital «Federico Gómez» who received treatment for the retention of several permanent teeth associated with CCD, avoiding extractions or prosthetics management. Treated with surgical exposure of the non-erupted teeth, the patient's occlusion was improved through forced eruption and orthodontic treatment. In this case report, emphasis has been given to the dental biomechanics used for performing the traction faster and achieving optimal results.

RESUMEN

La displasia cleidocraneal es una enfermedad poco frecuente y se caracteriza por la interrupción en el desarrollo óseo, la cual limita la exfoliación de los dientes deciduos, por lo que la dentición permanente no puede erupcionar y conlleva a afecciones estéticas y funcionales. El presente caso clínico describe a una paciente con displasia cleidocraneal atendida en el Hospital Infantil de México «Federico Gómez», la cual recibió tratamiento para varios dientes retenidos, asociados con la enfermedad de base, evitando la extracción de dientes incluidos y sin manejo protésico; tratada con exposición de los dientes incluidos, mejorando la oclusión por medio de exposición quirúrgica con tracción dental y tratamiento de ortodoncia. En este reporte de caso, se hace énfasis en la biomecánica utilizada para agilizar la tracción dental y obtener resultados óptimos.

Key words: Cleidocranial dysplasia, dental retentions, malocclusion, biomechanics.
Palabras clave: Displasia cleidocraneal, retenciones dentales, maloclusión, biomecánica.

BACKGROUND

Cleidocranial dysplasia (CCD) is a rare congenital skeletal disorder associated with clavicular hypoplasia or aplasia, delayed exfoliation of the temporal dentition and delayed eruption of permanent teeth.[1,2,4,6] The disorder is inherited as an autosomal dominant condition and 40% of cases occur spontaneously with no apparent genetic cause. The incidence is 1 per 1,000,000 inhabitants.

Every case of CCD present the following dentoalveolar characteristics to a greater or lesser degree:[2,5-7]

1. Retained deciduous teeth with reabsorbed roots.
2. Supernumerary teeth that displace permanent teeth and obstruct their eruption.
3. Delayed eruption due to low potential for eruption.
4. Reduced height of the lower third of the face, vertical growth of the alveolar bone is limited.
5. Late, but spontaneous eruption of permanent 1st and 2nd molars in both arches.
6. Severe delay in the development of the roots of the permanent teeth (approximately 3 years).

CASE DESCRIPTION

A female patient of 13 years and 10 months of age was admitted to the Orthodontic Service of the Children's Hospital of Mexico «Federico Gómez» with

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Mexico Children's Hospital «Federico Gómez».

diagnosis of cleidocranial dysplasia (CCD). The patient was skeletal Class I with a slightly reduced lower facial third. Intraoral examination showed a molar class I, incomplete permanent dentition, telescopic bite on the right side, 8 erupted permanent teeth in the maxillary arch and 9 permanent teeth in the mandibular arch, as well as deciduous teeth *(Figure 1)*.

During the functional examination, a tongue projection habit and a lisp were noticed when the patient spoke. These were caused by air leakage in the anterior area.

The panoramic radiograph showed 6 retained permanent teeth and agenesis of a lower incisor. The lateral headfilm showed a skeletal class I and proclined incisors, especially the upper ones *(Figure 2)*.

Treatment objectives

The objectives established in this treatment were: (1) to improve the pattern of eruption of retained permanent teeth by removing the deciduous teeth; (2) surgical exposure and traction of the retained permanent teeth; (3) improve facial and dental aesthetics; (4) restore dental and phonetic function; (5) facilitate future restorations; (6) establish optimal occlusal intercuspation.

Treatment alternatives

The main objective of dental treatment in patients with cleidocranial dysplasia is to achieve optimal

Figure 1.

Pretreatment facial and intraoral photographs. **A**, **B** and **C** correspond to the profile and frontal photographs at rest and smile. **D**, **E** and **F** correspond to right lateral, front and left lateral intraoral photographs. **G** and **H** correspond to upper and lower occlusal photographs respectively.

function, with the greatest number of natural teeth and aesthetic results that last into adulthood. The treatment alternatives considered in this case were:

1. **Extraction treatment.** Align both arches without closing spaces, remove all retained teeth and use the remaining teeth as support to provide a prosthetic solution.[1,2,7-9]
2. **No extraction treatment.** Expose the retained teeth and bond appliances that mediate the orthodontic traction of these to improve dental function and phonetics. At the end of the orthodontic treatment, the aesthetics would be improved by means of non-invasive restorations.[10-20]

The treatment options were explained to the parents under informed consent of the risks and benefits of each one of them. Treatment alternative two was chosen since it represented a conservative approach trying to pull the included teeth for subsequent alignment with orthodontics and thus avoid the use of dentures at an early age.[1,10,20,21]

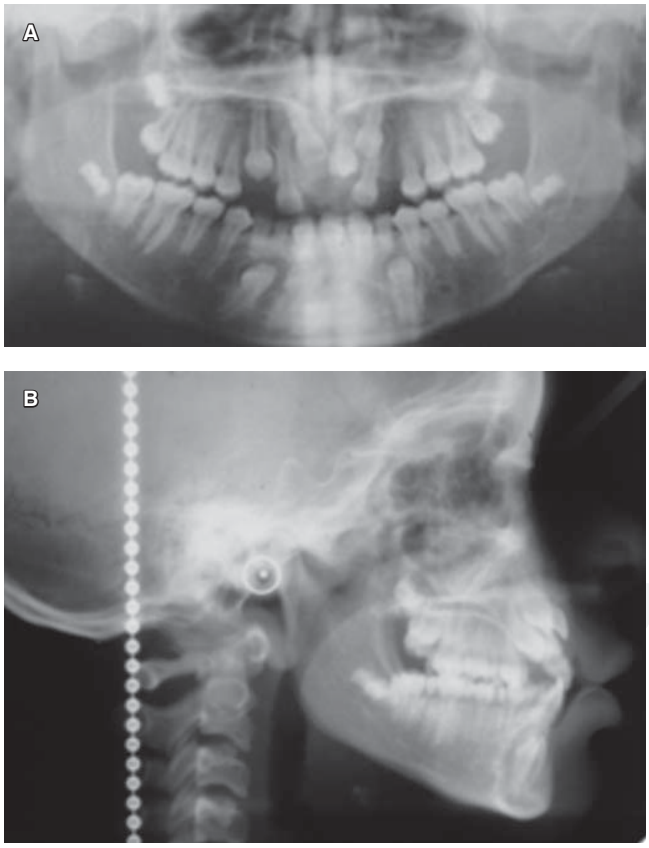

Figure 2. Pretreatment radiographs. **A** corresponds to the panoramic radiograph and **B** to the lateral headfilm.

Treatment progress

Phase 1. Tooth exposure and traction

Initially, in the first semester, the patient was referred to the Maxillofacial Surgery Service for the extraction of deciduous teeth. The next step was the exposure of the included teeth for bracket placement, regardless of prescription or position, with braided ligature that facilitated their traction.[20,22,24] A palatal arch was formed with welded hooks that allowed us to make activations to the metal ligature in order to pull each retained tooth until it was exposed to the oral cavity *(Figure 3)*.[10,14]

Six months later, devices for the bilateral traction of the retained canines were placed in the mandible, as well as lingual arches with welded hooks to improve traction direction[20,21,23] *(Figure 4).* In the maxilla, the retained dental organs were observed in the mouth so the palatal arch was removed and a 0.017" x 0.025" SS archwire was placed with a bypass in teeth #21 and 11, for the application of extrusive forces without repositioning brackets until the teeth adopted a more vertical position.

Phase II. Orthodontics

In the third semester, the upper teeth were in a better vertical position, so it was decided to perform bracket repositioning. The brackets of the upper left premolars were removed to allow the placement of a segmented arch with a gable bend that helped

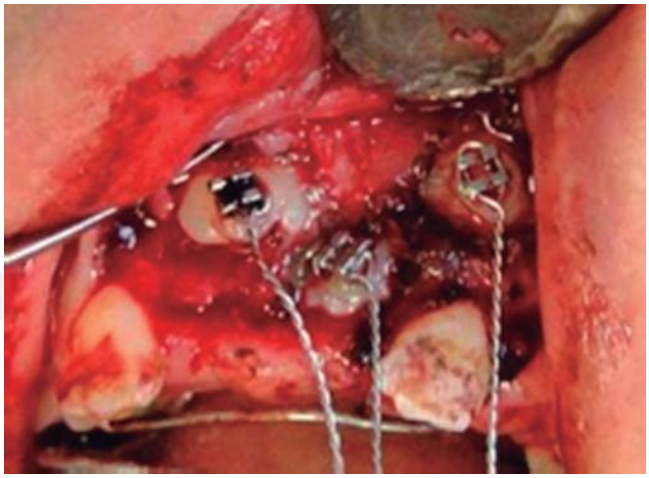

Figure 3. Surgical exposure of the retained teeth (Teeth #11, 21 and 23) and bracket placement with wire ligature for orthodontic traction. The palatal arch that aided the traction may be observed.

distalize the canine *(Figure 5)*. During this period, no changes in the lower arch were evident with the traction method of elastic modules tied to the lingual arch so it was removed and cantilevers (0.017" x 0.025" TMA) were placed at the cuspids with ligature wire (50-150 g).

During the fourth semester, after the monthly activation of the cantilevers (0.017" x 0.025" TMA) complete exposure of the lower canines was obtained.[14,22] Tooth #23 was distalized with the help of a segmented arch with double gable bends and all the fixed appliances used until now were removed to bond. 022" slot MBT brackets (3M Unitek, Monrovia, Cal.™). In the tooth #22 the bracket was placed at 180°, and the alignment and leveling of the arches with 0.014" CuNiti archwires was begun *(Figure 6)*.

We finished the fifth semester with 0.017" x 0.025" SS archwires conforming them to an Ovoid OrthoForm. No intermaxillary elastics were used and when the appliances were removed, teeth #11, 21 and the lower incisor were restored with resins. For the retention phase, circumferential Hawley retainers were used 24 hours a day for one year *(Figures 7 y 8)*.

RESULTS

Treatment was performed at the Orthodontic Service of the Children's Hospital of Mexico «Federico Gómez» during the period from April 2013 to April 2015.

The absence of anterior teeth was immediately treated when it was observed radiographically that

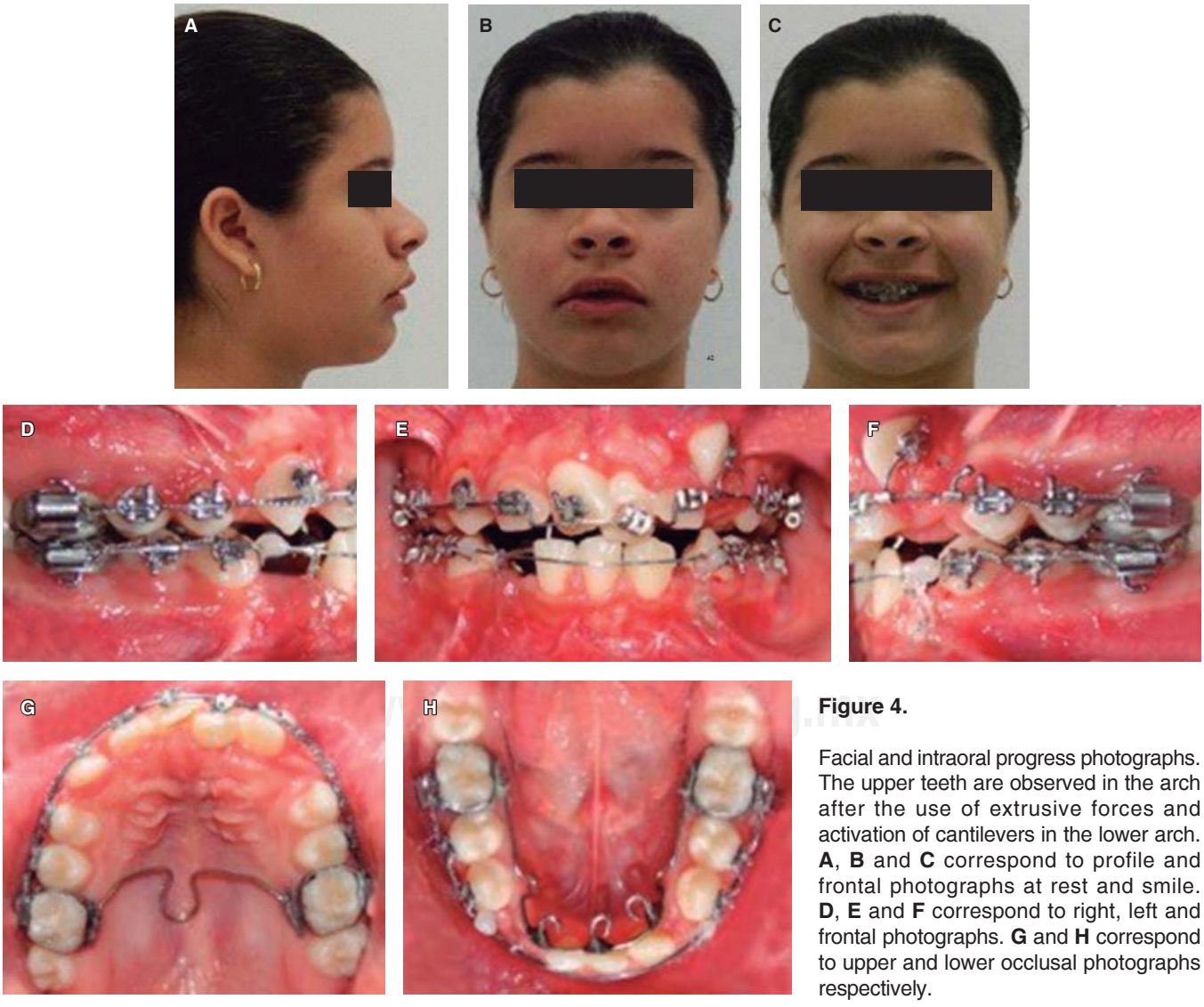

Figure 4.

Facial and intraoral progress photographs. The upper teeth are observed in the arch after the use of extrusive forces and activation of cantilevers in the lower arch. **A**, **B** and **C** correspond to profile and frontal photographs at rest and smile. **D**, **E** and **F** correspond to right, left and frontal photographs. **G** and **H** correspond to upper and lower occlusal photographs respectively.

root development of the incisors was complete without eruption. The Department of Maxillofacial Surgery was responsible for exposing the teeth and placing the brackets that helped to pull the permanent teeth.

The patient's dental conditions made the application of orthodontic forces efficient for the traction of several teeth without the need for extractions or the use of removable prostheses at a young age.

The early management of the edentulous area intercepted the habit of tongue projection and eliminated the list; there was an improvement in facial aesthetics, which helped with the patient's self-esteem. The final occlusion allowed good masticatory function.

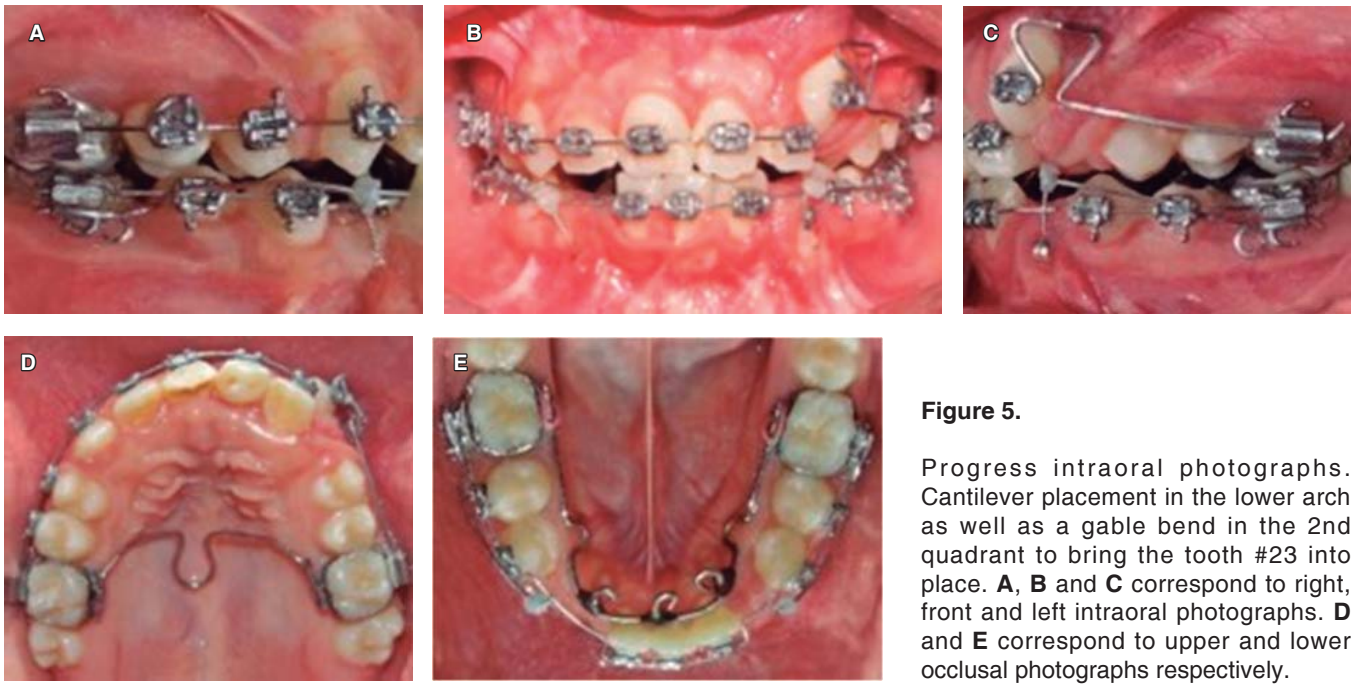

Figure 5.

Progress intraoral photographs. Cantilever placement in the lower arch as well as a gable bend in the 2nd quadrant to bring the tooth #23 into place. **A**, **B** and **C** correspond to right, front and left intraoral photographs. **D** and **E** correspond to upper and lower occlusal photographs respectively.

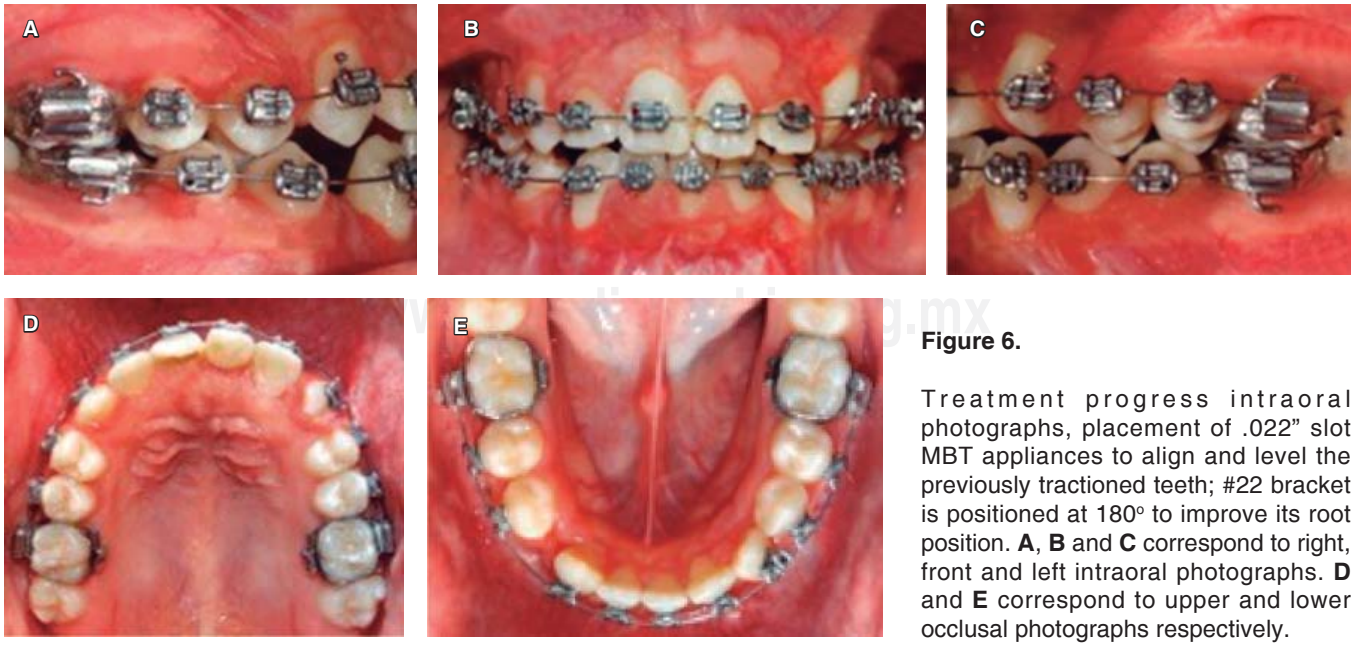

Figure 6.

Treatment progress intraoral photographs, placement of .022" slot MBT appliances to align and level the previously tractioned teeth; #22 bracket is positioned at 180° to improve its root position. **A**, **B** and **C** correspond to right, front and left intraoral photographs. **D** and **E** correspond to upper and lower occlusal photographs respectively.

DISCUSSION

The facial and dental characteristics of CCD have been described since 1911.[1,2,4] The afflicted patients commonly show disharmony between the dental arches, formation of supernumerary teeth and the potential lack of eruption of several permanent teeth.[4,6]

The treatment protocols described for patients with CCD such as those suggested by Becker, Douglas and Winter consider the surgical removal of the included teeth as a routine management for rehabilitation with removable prostheses, however, there are few cases described in young patients.

Gordon, in 1943 reports a study of dental characteristics in patients with CCD, which mentions that orthodontic treatment is unsatisfactory and discouraging and suggests the construction of removable prostheses.[2,3,13] Becker, recognizes the importance of early treatment of retained teeth.[14,15,19,22] In the case report hereby described, these factors were carefully considered and it was decided to begin dental traction as soon as possible and to leave detailed biomechanics for the orthodontic phase.[10,14]

The use of the lingual and palatal arches allowed a more labial and occlusal force vector, thus improving the dental position of the retained teeth.

The post-treatment appearance of the gingiva and the torque, alignment and position of the tractioned teeth in the arch were factors that determined treatment success.[14,15,17]

Using a combination of two treatment stages allowed us to successfully expose the clinical crowns

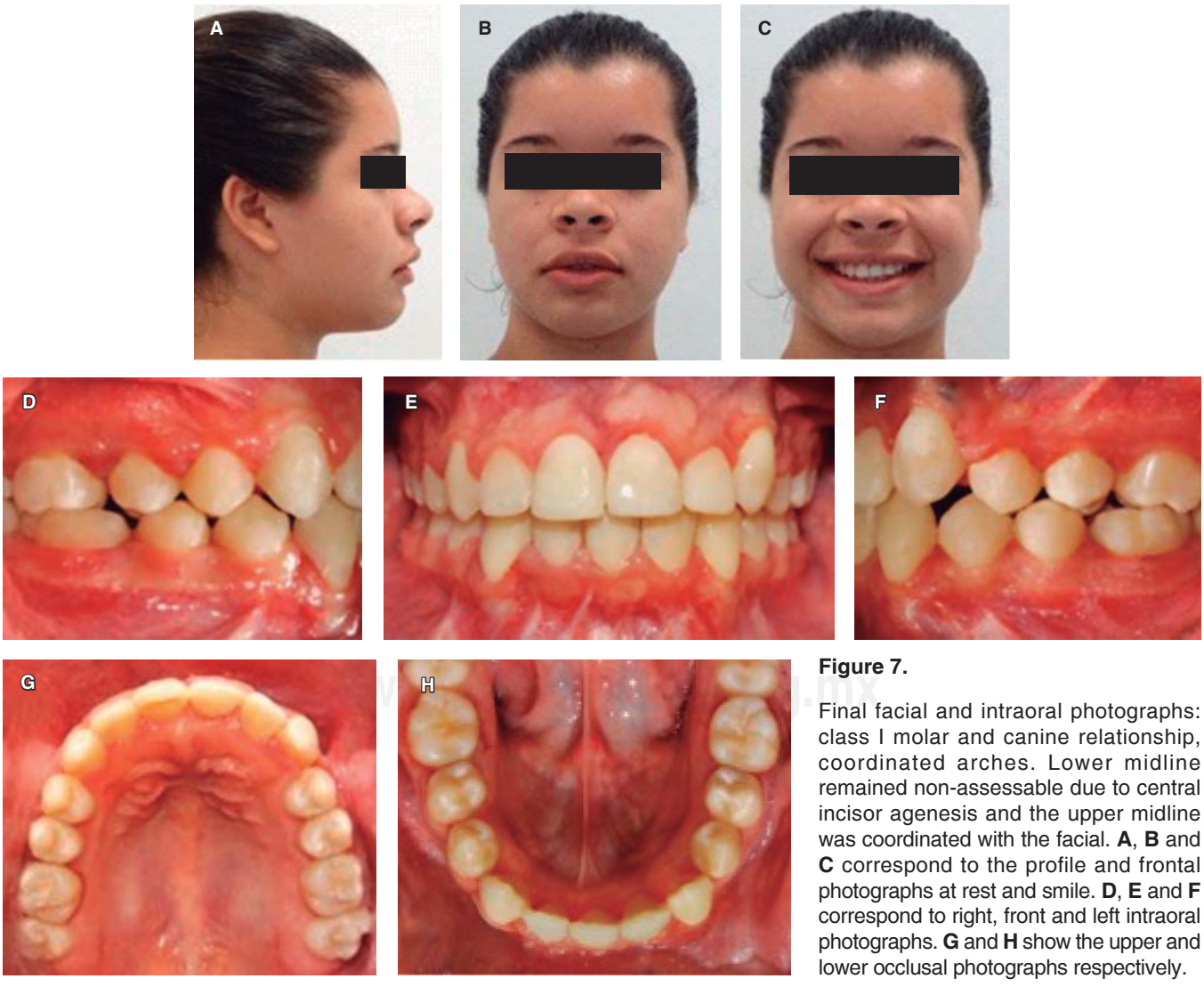

Figure 7.

Final facial and intraoral photographs: class I molar and canine relationship, coordinated arches. Lower midline remained non-assessable due to central incisor agenesis and the upper midline was coordinated with the facial. **A**, **B** and **C** correspond to the profile and frontal photographs at rest and smile. **D**, **E** and **F** correspond to right, front and left intraoral photographs. **G** and **H** show the upper and lower occlusal photographs respectively.

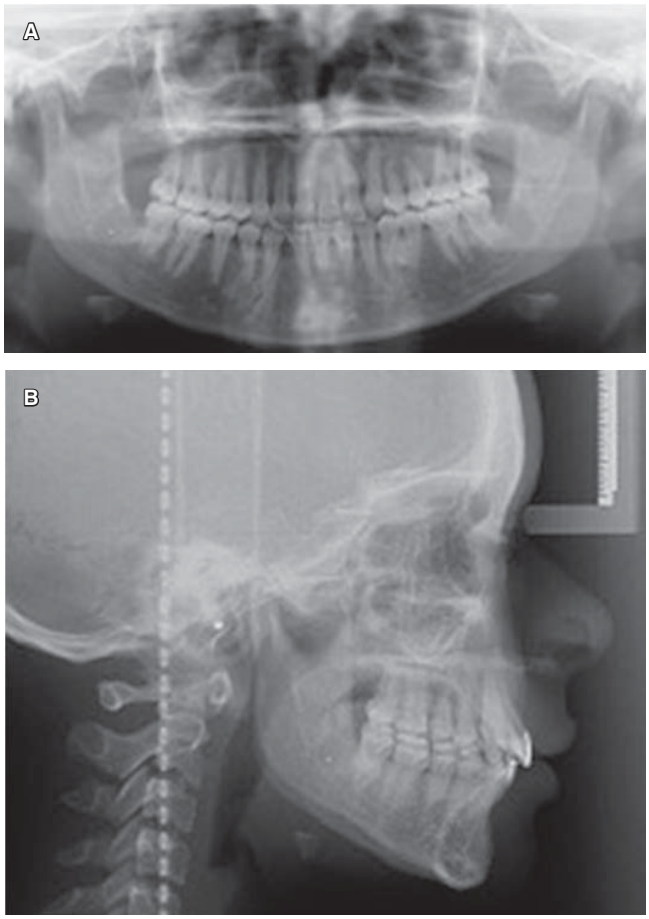

Figure 8. Postreatment radiographs. **A** corresponds to panoramic radiograph and **B** to the lateral headfilm.

of the included teeth and position them properly in the dental arches. The use of segmented arches studied and applied by Burstone, Becker and Pinho simplified the application of extrusive forces for the traction of the retained canines.[10,15,16,18,24]

The best age to treat eruptive problems is in the early stages, due to the maturity of the dental roots. However, failure may be due to ankylosis or external root resorption. The tensile forces must be light to prevent debonding of the fixed appliances, dental ankylosis, gingival recession or cant of the occlusal plane.[22-24]

The purpose of any dental treatment in patients with CCD is to provide masticatory function and improve the patient's appearance. To achieve these goals, several protocols have been developed and described in the literature, depending of the dentist's specialty, classifying them into three groups: (a) Prosthetic replacement by means of dentures with or without prior extractions of the impacted teeth. (b) A surgical approach involving the removal of supernumerary teeth, followed by replacement or transplantation of permanent teeth. (c) A combination of orthodontic surgical treatment aimed at forcing the eruption and aligning the permanent teeth.[24]

Once the diagnosis is determined, different treatment options are proposed to the patient, assessing different rehabilitation treatments to improve the aesthetics and including the improvement of the phonetic function.

The final occlusion of the patient was improved with the help of cantilevers. They were chosen for their ability to successfully maintain continuous and controlled forces (> 60 g) for teeth traction. Different elements were also used such as segmented archwires that showed excellent results in a short time.

For the fixed appliances, 0.022" slot MBT prescription was chosen because the authors, McLaughlin, Bennet and Trevisi, presented in their book the placement of the lateral incisor bracket at 180° as a characteristic of versatility, with the intention of improving the torque from +10 to -10° and promoting labial root movement as an option for palatally displaced lateral incisors.[26]

The Gable bend (0.017" x 0.025", TMA) was effective for distalization and control of upper left canine tipping. Optimal intercuspation was obtained after 10 months with MBT fixed appliances. The clinical difference was significant, dental traction induced vertical growth, correction of phonetics and aesthetics as well as functional balance of the patient.

CONCLUSIONS

The patient's function was restored without invasive therapy. Management of retained teeth in patients with CCD is a challenge for specialists. Treatment planning from a multidisciplinary perspective allows the outcome of treatment to be successful for the patient.

It is necessary to carefully consider the biomechanics since it is essential to improve the occlusion and facial aesthetics.

For our part, we suggest implementing dental traction without the need of invasive treatments such as extractions or prosthesis, protecting the patient's integrity while promoting the elimination of habits and an improvement in phonetics and aesthetics.

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